

Clinical Classification of Strength and Skill Impairments in Neurogenic Dysphagia

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Karen B. Ng, MA, BSc (Hons)

Department of Communication Disorders, University of Canterbury
The Rose Centre for Stroke Recovery and Research at St. George's Medical Centre
Christchurch, New Zealand
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Abstract

Swallowing is a complex, precise sequence of movements that is fundamentally mediated by brainstem mechanisms, but volitionally modulated by cortical processes. Although swallowing biomechanics are fairly well defined, little is known about the underlying pathophysiological mechanisms that lead to biomechanical impairments due to peripheral or central neurological damage. Behavioural rehabilitation approaches for dysphagia focus primarily on weakness as a presenting aetiology. This assumption is largely due to limitations in differential diagnosis of underlying pathophysiology. Research in the motor speech and limb literature suggests that peripheral muscle damage results primarily in weakness (decreased force generation), while central neurological lesions may be associated with impaired skill (decreased spatiotemporal precision of movement), with or without weakness. There is a need for accurate and specific diagnostic techniques in swallowing, so that improved and targeted rehabilitation strategies can be provided to patients with dysphagia. The aim of this research programme was to clinically classify patients with neurogenic dysphagia due to stroke or myopathy into subtypes based on objective measures of swallowing-related strength and movement precision.

Study 1 is a methodological study investigating the range of submental muscle activity utilised during minimum-, regular- and maximum-effort swallowing. Skill-based training and assessment protocols targeting movement precision in swallowing have used targets placed at submaximal levels of muscle contraction, to avoid any possible strengthening effects that might occur with maximum effort swallowing. Regular effort swallowing has decreased amplitude of submental surface electromyography (sEMG) activity compared to maximum effort swallowing. However, it is unknown whether, and to what extent, individuals can volitionally reduce magnitude of muscle contraction below that of regular effort swallowing. This information is important so that targets are not set below these minimal effort requirements. Forty-three healthy adults (22 female) representing four age groups (20-39, 40-59, 60-79, and 80+ years) participated in the study. They were verbally cued to swallow saliva and 5 mL water boluses using maximum, regular, and minimum levels of effort, in randomised order. Maximum peak amplitude and duration of each swallow were measured using sEMG. Results demonstrate that, on average, the minimum sEMG amplitude needed to generate functional swallowing was 31% of the participant's maximum swallowing muscle activity, suggesting that submaximal biofeedback targets used in skill-based protocols should not be set lower than this threshold. In addition, magnitude and duration of muscle activity during regular swallowing were more similar to minimum effort swallowing than maximum effort

swallowing, highlighting the possible functional relevance of submaximal swallowing over maximal effort swallowing in training paradigms.

In Study 2, healthy controls ($n=40$) and patients with dysphagia due to stroke ($n=55$) and myopathy ($n=19$) participated in a novel clinical assessment developed to differentiate between strength and movement precision impairments in swallowing. These groups were chosen because they were likely to have specific patterns of swallowing pathophysiology based on lesion location. They were assessed on the following four measures: swallowing strength, jaw-opening strength, swallowing movement precision, and jaw-opening movement precision. Submental muscle peak sEMG amplitude during effortful swallowing was utilised as a proxy for swallowing strength, while jaw-opening isometric strength was measured using dynamometry to provide comparative information from a volitional, non-swallowing task. Movement precision was measured as 1) hit rate, and 2) error in timing and force of submental muscle activation to place the peak of the sEMG signal in an on-screen target, during swallowing and non-swallowing jaw-opening tasks. Results indicated that compared to healthy controls, stroke patients demonstrated impaired performance on all strength and movement precision tests ($p < .01$) except for swallowing amplitude error, while patients with myopathy were impaired on strength tests only ($p < .01$). Hierarchical cluster analysis assigned participants to one of four clusters based on test performance. Cluster 1 contained primarily healthy controls and presented with better performance on strength and precision tests compared to other clusters. A second cluster comprised both myopathic and stroke patients, and was characterised by decreased performance on strength tasks but relatively intact precision. Stroke patients were further assigned to a third cluster (reduced performance in strength and jaw precision tests) or fourth cluster (deficits in strength and swallowing precision tests). Measures of movement precision were better able to classify participants into clusters, with prediction accuracy probabilities of 73 – 89%, compared to strength measures. Results from this study reveal that several subtypes of swallowing pathophysiology may be identified after stroke. It is possible that these different clusters of patients with dysphagia would benefit from different rehabilitation approaches. The novel clinical assessment was able to differentiate between groups that were expected to have distinct patterns of strength and movement precision impairments, suggesting potential as an adjunct clinical test for differential diagnosis of underlying pathophysiology. Assessment of movement precision in swallowing may be an important but overlooked aspect of rehabilitation that should be further explored in controlled studies.

In clinical practice, measures of hyoid movement on videofluoroscopic swallowing studies (VFSS) are sometimes used to make judgements about swallowing strength, although the relationship between biomechanical measures of hyoid movement and underlying pathophysiology are unclear. Study 3 was an initial attempt to explore this relationship. Eight stroke patients who participated in the novel strength and movement precision assessment in Study 2 also participated in a swallowing assessment of 5 mL liquid and 5 mL puree using VFSS. Biomechanical measures of hyoid displacement, hyoid burst duration, hyoid burst velocity, and stage transition duration obtained from VFSS were correlated with physiological measures of strength and movement precision from dynamometry and sEMG. Overall, hyoid trajectory was not specifically associated with measures of strength nor movement precision, suggesting that there are many complex factors contributing towards hyoid movement, and it may be difficult to infer physiology from visualisation of hyoid movement on VFSS.

Results of these research studies challenge the prevailing assumption that reduced force generation is the predominant cause of dysphagia, particularly after central neurological damage such as stroke. This thesis identifies the presence of several subgroups in patients with post-stroke dysphagia, and provides characterisation of each subgroup's impairment patterns. We propose that decreased movement precision is one of the possible underlying mechanisms of swallowing impairment that may co-occur with weakness. Initial feasibility of a clinical assessment of strength and movement precision has been established, setting the framework for further research in the physiological, rather than the purely biomechanical, assessment of dysphagia. Improving the accuracy and specificity of diagnosis of swallowing pathophysiology is fundamental to the effective management of dysphagia.

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This PhD thesis conforms to the referencing style described in the Publication Manual of the American Psychological Association (6th ed.), and follows spelling recommended by the Oxford Dictionaries (www.oxforddictionaries.com).

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Conference presentations

- European Society of Swallowing Disorders Annual Congress (2018; Dublin, Ireland)
- Stroke Rehab: From No-Tech to Go-Tech (2018; Christchurch, New Zealand)
- University of Canterbury Postgraduate Showcase (2017; Christchurch, New Zealand)
– awarded 1st place, and joint winner for People's Choice Award
- Dysphagia Research Society Annual Meeting (2017; Portland, USA)
- University of Canterbury Thesis in Three Competition (2016; Christchurch, New Zealand) – awarded 1st place in PhD category
- Biomouth Symposium (2016; Lincoln, New Zealand)
- University of Canterbury Postgraduate Showcase (2015; Christchurch, New Zealand)
– awarded 2nd place

List of Abbreviations

1-RM	1-repetition maximum
ANOVA	Analysis of variance
BA	Brodmann's area
BiSSkiT	Biofeedback in Strength and Skill Training
BG	Basal ganglia
C1	Cervical spine nerve 1
C2	Cervical spine nerve 2
C3	Cervical spine nerve 3
C4	Cervical spine nerve 4
CI	Confidence interval
CN	Cranial nerve
CNS	Central nervous system
CPG	Central pattern generator
CSA	Cross-sectional area
DSG	Dorsal swallowing group
EAT-10	10-Item Eating Assessment Tool
EMG	Electromyography
EMST	Expiratory muscle strength training
ES	Effortful swallowing
FEES	Fibrendoscopic evaluation of swallowing
fMRI	Functional magnetic resonance imaging
IBM	Inclusion body myositis
ICC	Intra-class correlation coefficient
JAE	Jaw amplitude error
JF	Jaw force
JOFT	Jaw-opening force test

JTE	Jaw temporal error
LMN	Lower motor neuron
M1	Motor strip, Brodmann's area 4
MANOVA	Multivariate analysis of variance
MD	Muscular dystrophy
MRI	Magnetic resonance imaging
MVC	Maximum voluntary contraction
N	Newtons
NA	Nucleus ambiguous
NG	Nasogastric
NTS	Nucleus tractus solitaries
PEG	Percutaneous endoscopic gastrostomy
PES	Pharyngoesophageal segment
rTMS	Repetitive transcranial magnetic stimulation
S1	Sensory strip, Brodmann's area 3, 1, 2
SAE	Swallowing amplitude error
SCI	Spinal cord injury
SD	Standard deviation
sEMG	Surface electromyography
SHR	Swallowing hit rate
SLP	Speech-language pathologist
SMA	Supplementary motor area
STD	Stage transition duration
STE	Swallowing temporal error
SWAL-QOL	Swallowing quality of life outcome tool
TOMASS	Test of Masticating and Swallowing Solids
TWST	Timed Water Swallowing Test

UES	Upper oesophageal sphincter
UMN	Upper motor neuron
VFSS	Videofluoroscopic swallowing study
VIF	Variance inflation factor
VSG	Ventral swallowing group

PART A. INTRODUCTION AND LITERATURE REVIEW

Chapter 1. Introduction

Dysphagia is a common symptom of neurological disease, and is associated with serious medical, social, and emotional consequences. There is a need for the development of assessment techniques that can identify the underlying mechanisms of swallowing impairment, so that appropriate treatment protocols can be selected to address the deficits. However, there has historically been a presumption of weakness as the primary cause of dysphagia. This is likely due, in part, to assessment tools that lack the specificity to differentiate between biomechanical impairment caused by decreased force generation, decreased movement precision, and/or other abnormalities. Both decreased strength and decreased movement precision can cause similar biomechanical impairments, making it difficult to identify the underlying cause. Studies in the limb and motor speech literature, as well as emerging research in the field of dysphagia, suggest that motor impairments in swallowing may be classified by different patterns of underlying impairment. Peripheral lesions are characterised by weakness or decreased force generation. Central lesions may be typified by reduced coordination and poor movement precision, with or without concurrent weakness. This research programme refines methods used for measuring strength and movement precision in swallowing, investigated the viability of a strength and movement precision assessment, and explored the relationship between physiological and biomechanical measures of swallowing.

Part A provides an in-depth review of the literature. In Chapters 2 and 3, normal and disordered swallowing are examined, with a particular emphasis on the neural control of swallowing, the pathophysiological mechanisms underlying neurogenic dysphagia, and the strengths and limitations of current assessment methods. Chapter 4 focuses on two particular pathophysiological features, impaired strength and decreased movement precision. Since there is limited research comparing strength and movement precision in swallowing, findings from the limb and motor speech literature are first reviewed to glean some possible insights into swallowing behaviour. Evidence for the influence of strength and movement precision impairments on functional limb, motor speech, and swallowing behaviour is provided, as well as current methods of quantifying and discriminating between the two impairments.

In Part B, three experimental studies are described. Chapter 5 provides an overview of the objectives and hypotheses for these studies. Chapter 6 presents Study 1, a methodological study that measured the minimum to maximum range of muscle activity used by healthy adults during functional swallowing. This knowledge is important for rehabilitation protocols that use surface electromyographic (sEMG) biofeedback, so that targets are placed in a range that is

physiologically achievable. Knowledge of the upper limit is needed for strength-based training that focuses on increasing maximal muscle contraction, while the lower limit is needed for skill-based training protocols that target precise control and gradation of submaximal muscle activation. Besides answering a methodological query that contributes to Study 2, this study also provides insight into the minimum threshold and relative proportion of muscle activity needed for swallowing in healthy participants, which may have implications on rehabilitation methods.

Chapter 7 details Study 2, the main study of the research programme. This study was inspired by the desire to further define the underlying pathophysiological mechanisms of dysphagia. While there are likely many pathophysiological features underlying dysphagia, this study represents the first step toward systematically investigating these features by focusing on strength and a broader category of movement precision. The study explored the ability of a prototype clinical assessment to classify individuals based on patterns of test performance. The three groups of participants (healthy controls, patients with dysphagia after stroke, and patients with dysphagia due to myopathy) were chosen to participate in the assessment based on their expected patterns of swallowing performance. Given the lack of research on movement precision, several measures were used to explore the sensitivity of the novel assessment. The chapter discusses the results, and the theoretical and clinical implications of the study. The ability to differentiate between weakness and impaired movement precision in patients with dysphagia will challenge the historical assumption of weakness and provide evidence for future development of improved assessments and impairment-specific treatments.

As described in Chapter 8, VFSS is commonly used in clinical practice to identify swallowing impairments, with weakness often being assumed to be the cause of impaired biomechanical movement. However, the relationship between weakness and abnormal hyoid movement is unclear, and deficits in movement precision may also contribute to impaired biomechanical movement. Study 3 investigated the relationships between biomechanical measures from VFSS and physiological measures from the novel strength and movement precision assessment. Since there has not been previous research on the effects of movement precision on biomechanical movement, this study was an exploratory first step in understanding the association between biomechanical impairments and underlying pathophysiology.

In Chapter 9, the combined results of the three studies are discussed, and clinical implications of the results are offered. In particular, the importance of questioning underlying assumptions regarding swallowing pathophysiology is emphasised. It is only by systematically investigating

these long-held beliefs, that accurate and specific management techniques can be developed. This research programme is the first to propose a clinical assessment to differentiate between strength and movement precision in swallowing, and provide objective data from patients with dysphagia and healthy controls, paving the way for further investigations.

Chapter 2. Normal Swallowing

2.1 Swallowing physiology

Swallowing is a complex, stereotyped behaviour involving the sequential activation and inhibition of bilateral nerves and muscles in the mouth, pharynx, larynx and oesophagus (Jean, 2001). This sensorimotor response occurs rapidly, usually within the span of 0.6 – 1 s, but is highly coordinated (Ertekin & Aydoğdu, 2003). Safe and efficient swallowing relies not only on adequate muscle contraction and force, but the integration of sensory feedback and descending cortical input, as well as the precise timing and amplitude of motor responses.

Swallowing serves two life-sustaining functions: propelling food and liquid from the oral cavity into the stomach and thus fulfilling nutritional and hydration needs, and protecting the upper respiratory tract from the invasion of foreign material (Jean, 2001). Swallowing historically has been described as having three phases: oral, pharyngeal, and oesophageal (Ertekin & Aydoğdu, 2003), however the increasingly important role of cognition and sensory input in swallowing has justified the inclusion of a pre-oral phase (Leopold & Kagel, 1997). Regardless of the model used to conceptualise swallowing, it is important to note that the phases do not operate in isolation, as they overlap in time and are functionally interdependent (Martin-Harris, Michel, & Castell, 2005).

2.1.1 *Pre-oral phase*

Before the bolus arrives in the oral cavity, visual and olfactory stimuli from the bolus activate the cerebral cortex via sensory receptors from the optic and olfactory cranial nerves (Maeda et al., 2004). This visual and olfactory input prepares the swallowing system for the oncoming bolus and modifies subsequent swallowing phases. Other factors such as hunger, emotion, attention, and cognition also play a role in this pre-oral cognitive or anticipatory phase (Leopold & Kagel, 1997). Presentation of a meal may stimulate the glossopharyngeal and facial nerves, initiating the secretion of saliva from the submandibular, sublingual, and parotid glands. Saliva aids in digestion, as well as bolus formation and transport (Pedersen, Bardow, Jensen, & Nauntofte, 2002). Salivary secretion can even be initiated by imagining food, illustrating the effect of cognition and descending cortical inputs on swallowing behaviour (Pedersen et al., 2002). The modulatory effect of olfactory stimuli prior to ingestion was demonstrated by Ebihara et al. (2006), who showed that inhaling black pepper oil not only modified subsequent swallowing behaviour, but also activated neurophysiological changes in

the insular and orbitofrontal cortices. In another study, drink-related visual inputs (e.g., photograph of a glass of beer) presented before swallowing shortened the latency of water swallowing onset, highlighting the modulatory effect of pre-oral supramedullary inputs on ingestive swallowing (Maeda et al., 2004).

2.1.2 Oral phase

The oral phase begins when the bolus reaches the oral cavity. The orbicularis oris muscle, which encircles the lips and is innervated by the buccal branches of the facial nerves, is inhibited to allow the lips to open and accept the oncoming bolus. Other facial muscles innervated by the buccal and zygomatic branches of the facial nerves are activated to pull lips superiorly and laterally for acceptance of larger boluses, namely the risorius, zygomatic major, levator labii superioris, and levator anguli oris muscles (Daniels & Huckabee, 2013; Perlman & Christensen, 2003). Jaw opening is achieved by contraction of the submental muscles. These include the anterior bellies of the digastric and mylohyoid muscles, which are innervated by the mandibular branches of the trigeminal nerves, and geniohyoid muscles, innervated by the ansa cervicalis (Perlman & Christensen, 2003). The submental muscles elevate the hyoid if the jaw is fixed, and open the jaw if the hyoid is fixed.

After the bolus has entered the oral cavity, it is contained there by the tongue, lips, and hard palate. Contraction of the palatoglossus muscles (innervated by the pharyngeal plexus) results in approximation of the base of tongue and the soft palate; this glossopalatal seal maintains the bolus in the oral cavity to avoid premature spillage into the pharynx (Perlman & Christensen, 2003). The styloglossus muscles contribute to glossopalatal seal by drawing the tongue upwards and backwards towards the palate.

The hypoglossal nerves control all intrinsic and most extrinsic muscles of the tongue, and hence are responsible for changing the surface contour and position of the tongue for bolus containment and manipulation (Perlman & Christensen, 2003). The intrinsic lingual muscles (superior longitudinal, inferior longitudinal, transverse, and verticalis) alter the tongue surface to accept the bolus, drop the tongue midline to contain the bolus, and elevate the midline to allow the bolus to reach the teeth for mastication (Daniels & Huckabee, 2014). The extrinsic muscles (hyoglossus, genioglossus, and styloglossus) move the tongue within the oral cavity to collect the bolus. Sensory information from the oral cavity and oropharynx, carried by the trigeminal and glossopharyngeal nerves to the brainstem and cortex, facilitates motor planning and execution for bolus manipulation (Fuller et al., 2012; Perlman & Christensen, 2003). Motor

information carried by the trigeminal nerves to the muscles of mastication (masseter, temporalis, lateral, and medial pterygoid muscles) allows for rotary and lateral movement of the mandible when chewing. Cyclic and rhythmic jaw movements are spatially and temporally coordinated with movements of the tongue, cheek, and soft palate (Matsuo & Palmer, 2009).

The onsets of apnoea (cessation of breathing), vocal fold closure, and arytenoid adduction often occur prior to, or in close temporal relation to, oral transfer of the bolus to the pharynx (Hiss, Strauss, Treole, Stuart, & Boutilier, 2004; Shaker, Dodds, Dantas, Hogan, & Arndorfer, 1990; Van Daele, McCulloch, Palmer, & Langmore, 2005). When the bolus is prepared for transfer into the pharynx, activation of the pharyngeal plexus is terminated, causing the palatoglossus muscles to relax and the tongue base to descend to resting position. The genioglossus and hyoglossus muscles (innervated by the hypoglossal nerves) are activated, pulling the midline of the tongue towards the palate anteriorly to posteriorly in a wavelike fashion to transfer the bolus out of the oral cavity (Kahrilas, Lin, Logemann, Ergun, & Facchini, 1993). The end of the oral phase is often marked by the bolus reaching the ramus of the mandible.

2.1.3 Pharyngeal phase

The nucleus tractus solitarius (NTS) in the brainstem receives sensory information about the bolus via the trigeminal, facial, and glossopharyngeal nerves, as well as cognitive input from the cortex. When the input reaches a certain threshold, the patterned motor response of a pharyngeal swallow is triggered (Ertekin, 2011; Ertekin & Aydoğdu, 2003). Initiation of pharyngeal swallowing usually occurs when the head of the bolus reaches a certain location, e.g., the ramus of the mandible (Logemann, 1998), although many healthy individuals demonstrate pharyngeal swallowing onset after the bolus has already passed the mandible (Martin-Harris, Brodsky, Michel, Lee, & Walters, 2007). Normal variation in pharyngeal phase initiation can depend on many factors including age (Logemann et al., 2000), bolus type (Robbins, Hamilton, Lof, & Kempster, 1992), and method of bolus administration (e.g., cup or straw; Daniels et al., 2004).

Once the pharyngeal swallowing response is triggered, the fast and overlapping activation of cranial nerves and muscles results in the subsequent cascade of swallowing events. The levator veli palatini muscles elevate the soft palate for velopharyngeal closure. This muscle is innervated by the pharyngeal plexus, which is made up of pharyngeal branch fibres of the glossopharyngeal and vagus nerves (Kitagawa, Shingai, Takahashi, & Yamada, 2002). Velopharyngeal closure makes a seal between the oropharynx and nasopharynx, contributing

to increased pharyngeal pressure during swallowing (Daniels & Huckabee, 2014; Logemann, 1998).

The submental muscles (mylohyoid, geniohyoid, and anterior bellies of the digastric muscles) in combination with the posterior digastric and stylohyoid muscles, form the suprahyoid muscles (Figure 2.1). This muscle group contributes to anterior and posterior movement, as well as elevation of the hyolaryngeal complex (Pearson, Hindson, Langmore, & Zumwalt, 2013). The larynx and hyoid bone are pulled superiorly and anteriorly by excitation of the trigeminal nerves, which activates the anterior bellies of the digastric and mylohyoid muscles, and the ansa cervicalis, which innervates the geniohyoid muscles (Pearson, Langmore, Yu, & Zumwalt, 2012). Anterior hyolaryngeal movement provides traction forces to pull open the upper oesophageal sphincter (UES; Cook et al., 1989) and facilitates epiglottic deflection for airway protection (Vandaele, Perlman, & Cassell, 1995). Superior movement of the hyolaryngeal complex further contributes to supraglottic closure (Logemann, 1998).

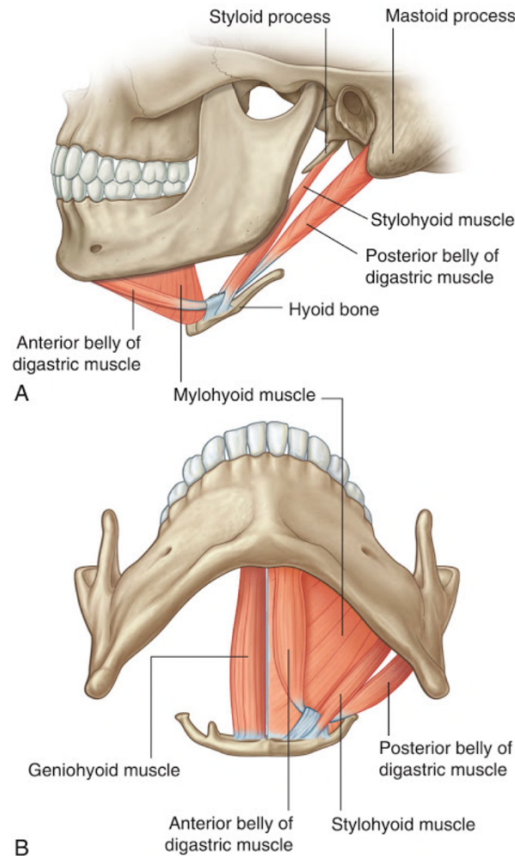


Figure 2.1. Suprahyoid muscles. A: Lateral view; B: inferior view. From “Gray’s Anatomy for Students E-Book” 3rd Ed., by R. Drake, A. W. Vogl, and A. W. M. Mitchell, p. 1008. Copyright 2014 by Elsevier. Reprinted with permission.

Laryngeal closure can be described in three components: glottic closure of the true vocal folds, closure of the ventricular (false) vocal folds associated with approximation of the arytenoids and base of epiglottis, and epiglottic inversion (Inamoto et al., 2011; Logemann et al., 1992). The vagus nerves are critically important for closure of the laryngeal vestibule and opening of the UES, allowing the bolus to be directed away from the airway and into the oesophagus. Contraction of the bilateral interarytenoid, lateral cricoarytenoid, and thyroarytenoid muscles result in vocal fold adduction prior to and during swallowing (Fuller et al., 2012; Perlman & Christensen, 2003). The sensory mechanisms of airway protection also rely heavily on the vagus nerves. The recurrent laryngeal nerves of the vagus are critical for subglottic sensation, and the internal branches of the superior laryngeal nerves of the vagus carry sensory information from the supraglottis (Erman, Kejner, Hogikyan, & Feldman, 2009). Detection of any foreign material in the laryngeal vestibule by vagus nerve endings results in signals sent to both the sensory and motor nuclei in the brainstem, allowing for a cough to be produced and foreign material expelled immediately (Carr, 2004).

Bolus propulsion through the pharynx is accomplished by excitation of the facial and hypoglossal nerves and the pharyngeal plexus. The driving pressure of the base of tongue on the bolus tail and contact between the tongue base and posterior pharyngeal wall propels the bolus posteriorly and inferiorly through the pharynx (Kahrilas, Logemann, Lin, & Ergun, 1992). Tongue base retraction and depression is facilitated by contraction of the posterior bellies of the digastric and stylohyoid muscles (digastric branches of the facial nerves), hyoglossus and genioglossus muscles (hypoglossal nerves), and glossopharyngeus muscles (pharyngeal plexus). The glossopharyngeus, along with the salpingopharyngeus and palatopharyngeus muscles, also contribute to pharyngeal shortening. This shortening decreases the distance the bolus has to travel and builds intraluminal pressure (Palmer, Tanaka, & Ensrud, 2000). Finally, the pharyngeal plexus activates the superior, middle, and inferior constrictor muscles sequentially with circular contraction of the pharynx to clear pharyngeal residuals (Daniels & Huckabee, 2014; Kahrilas et al., 1992).

In order for the bolus to pass from the pharynx to the oesophagus, it must enter the tonically-contracted UES. This occurs in a coordinated sequence of events, firstly through relaxation of the cricopharyngeus muscle, and then anterior movement of the hyolaryngeal complex which pulls the relaxed UES open. Inhibition of fibres from the recurrent laryngeal nerves and external branches of the superior laryngeal nerves allows for relaxation of the cricopharyngeus muscle. Excitation of the trigeminal nerves then contracts the anterior bellies of the digastric and mylohyoid muscles, and activation of the ansa cervicalis contracts the geniohyoid muscles, resulting in anterior hyolaryngeal excursion which pulls the relaxed UES open. As the bolus enters the sphincter, intrabolus pressure can further widen the UES opening (Cook et al., 1989).

Swallowing and respiration share a common pathway – the aerodigestive tract. During swallowing, there is a very short period of apnoea when the usual phases of respiration (inhalation and exhalation) are interrupted. Once pharyngeal swallowing is completed, an open laryngeal airway is re-established and respiration can continue. Respiratory and physiologic swallowing events are highly temporally coordinated to allow for adequate exchange of gasses and to prevent aspiration of material into the airway (Martin-Harris, Brodsky, Price, Michel, & Walters, 2003). Most healthy individuals demonstrate a respiratory-phase pattern of expiration immediately before, and after, swallowing, although there is some normal variation. For example, approximately 20% of healthy individuals swallow on inhalation (Martin-Harris, Brodsky, et al., 2005).

2.1.4 Oesophageal phase

The oesophagus is a muscular tube that contains striated and smooth muscle. The bolus enters the top of the oesophagus at the UES, and is pushed through the oesophagus by a sequential peristaltic wave until it passes through the lower oesophageal sphincter and into the stomach (Goyal & Chaudhury, 2008). Bolus transport through the oesophagus can be described in four phases (Miller et al., 1995). Before the bolus arrives, the oesophagus is in phase 1 (resting), with no luminal opening. The oesophagus then passively stretches open upon the arrival of the bolus in phase 2 (passive distension), and demonstrates maximum peak pressure at phase 3 (contraction). Finally, phase 4 (relaxation) sees the oesophagus return to baseline lumen circumference and intraoesophageal pressure (Miller et al., 1995).

2.2 Neural control of swallowing

The neural control of swallowing reflects the complexity of this patterned motor response. Swallowing is mediated by a distributed neural network, involving the interaction and coordination of systems across multiple levels of the neural axis (cortical, subcortical, brainstem, and peripheral; Leopold & Daniels, 2010). Descending inputs from cortical and subcortical structures, as well as sensory afferent input from cranial nerves, converge on a swallowing central pattern generator (CPG) and cranial nerve nuclei in the brainstem, subsequently activating oral, pharyngeal, and oesophageal muscles and triggering a swallowing response (Martin & Sessle, 1993). The extent and duration of movement during swallowing can be mediated by volitional control (Humbert & German, 2013), e.g., during voluntary swallowing manoeuvres (Wheeler-Hegland, Rosenbek, & Sapienza, 2008). Given the complexity and wide distribution of the neural control of swallowing, damage at any location of the neural axis can cause a range of swallowing deficits.

2.2.1 Brainstem and peripheral control of swallowing

The brainstem and cranial nerves are essential for initiating and controlling the basic sequence of swallowing. Of the 12 cranial nerves, five pairs are heavily involved in swallowing control: the trigeminal, facial, glossopharyngeal, vagus, and hypoglossal cranial nerves. These cranial nerves are crucial in relaying information between the brainstem swallowing centres and the oropharyngeal structures responsible for swallowing. The afferent cranial nerves transmit sensory information (including touch, pain, temperature, and taste) from peripheral sensory receptors in the mouth, pharynx, and larynx to the brainstem nuclei located in the central

nervous system. The efferent cranial nerves emerge from the brainstem nuclei and innervate the peripheral muscles (Ertekin & Aydoğdu, 2003).

Pharyngeal swallowing can be elicited even when cortical and subcortical structures are removed above the level of the pons and medulla, indicating the importance of brainstem regions (Miller, 1999). The presence of a “swallowing centre” in the brainstem that controls deglutition was investigated by Doty et al. (1967), who used electromyographic (EMG) methods to show that a functional grouping of interconnected neurons in the medulla were responsible for producing this automatic sequence. Similar to CPGs in respiration and locomotion, this swallowing CPG is a circuit of motoneurons and interneurons that produces a rhythmic and sequential motor pattern even in the absence of sensory feedback (Jean, 2001; Marder & Bucher, 2001).

The swallowing CPG (Figure 2.2) consists of two main groups of bilateral neurons located in the medulla: the dorsal swallowing group (DSG) located within and around the nucleus tractus solitarius (NTS) and adjacent reticular formation, and the ventral swallowing group (VSG) located in the ventrolateral medulla (VLM) adjacent to the nucleus ambiguus (NA). The DSG receives afferent inputs from the periphery (for example, oropharyngeal receptors during eating and drinking) and from supramedullary structures like the cortex (Ertekin, 2011; Ertekin & Aydoğdu, 2003). Generator neurons in the DSG initiate and shape the swallowing pattern, and, in turn, activate neurons in the VSG. The premotor neurons of the VSG then drive the motoneuron pools of cranial and cervical spinal nerves involved in swallowing (trigeminal, facial, glossopharyngeal, vagus, and hypoglossal cranial nerves and cervical spinal nerves 1-3; Jean, 2001).

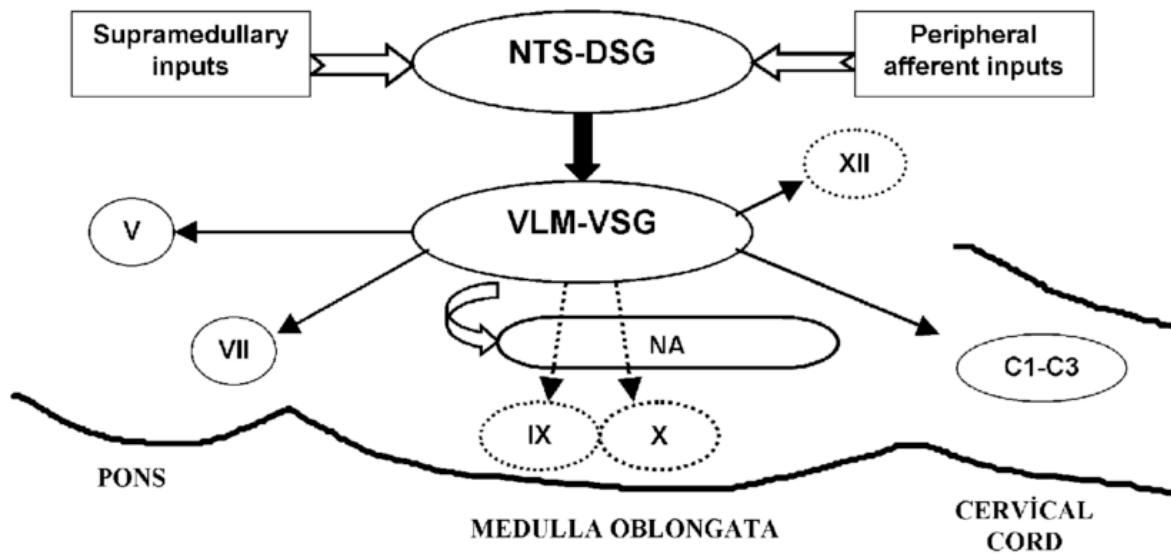


Figure 2.2. Schematic diagram of the swallowing central pattern generator (CPG) in the brainstem. The dorsal swallowing group (DSG) receives supramedullary and peripheral inputs, initiates and programs the swallowing response, and activates the ventral swallowing group (VSG). The VSG distributes the rhythmic swallowing response to motor neurons involved in swallowing. From “Neurophysiology of Swallowing,” by C. Ertekin and I. Aydogdu, *Clinical Neurophysiology*, Vol. 114, p. 2234. Copyright 2003 by Elsevier. Reprinted with permission.

2.2.2 Supranuclear control of swallowing

Historically, studies have focused on brainstem control of swallowing. However, research has evolved over time to now recognise the crucial role the supranuclear regions play in the regulation of swallowing (Miller, 2008). Swallowing impairment after cortical and subcortical damage such as stroke is a widely-recognised clinical problem, and thus it is important to understand the underlying cortical mechanisms of swallowing. Given the crucial role the brainstem plays in the initiation and control of swallowing, it was traditionally thought that swallowing impairments could only occur after brainstem or bilateral cortical lesions (Meadows, 1973). However, lesion studies have provided evidence that unilateral cortical and/or subcortical lesions can often cause dysphagia, supporting the idea that supramedullary structures have an important role in swallowing (Daniels & Foundas, 1999; Daniels, Foundas, Iglesia, & Sullivan, 1996; Martin & Sessle, 1993). Primate studies have also demonstrated cortical control of swallowing. Swallowing behaviours, as verified by EMG activity, can be elicited in awake primates using intracranial microelectrode stimulation in the face sensorimotor cortices (Martin et al., 1999), and lesions of the lateral precentral gyrus in

monkeys and anterolateral frontal cortex in rabbits can impair mastication and swallowing (Martin & Sessle, 1993).

Advancements in neuroimaging techniques have provided further evidence and quantification of the role of the cortex in swallowing. Functional magnetic resonance imaging (fMRI) studies in humans have showed multifocal and bilateral cortical activity during both automatic and volitional swallowing (Figure 2.3; Hamdy et al., 1999; Martin, Goodyear, Gati, & Menon, 2001). Cortical areas frequently found to be involved in swallowing include the sensorimotor cortex, prefrontal cortex, supplementary motor area, anterior cingulate gyrus, insula, parietooccipital area, and temporal cortex (Ertekin & Aydoğdu, 2003; Malandraki, Sutton, Perlman, Karampinos, & Conway, 2009; Michou & Hamdy, 2009). Many years later, Verin, Michou, Leroi, Hamdy, & Marie (2012) investigated the effect of a “virtual” lesion of the cortex on healthy swallowing. They found that using repetitive transcranial magnetic stimulation (rTMS) to inhibit the oropharyngeal motor cortex resulted in a significant change in videofluoroscopic bolus flow measurements.

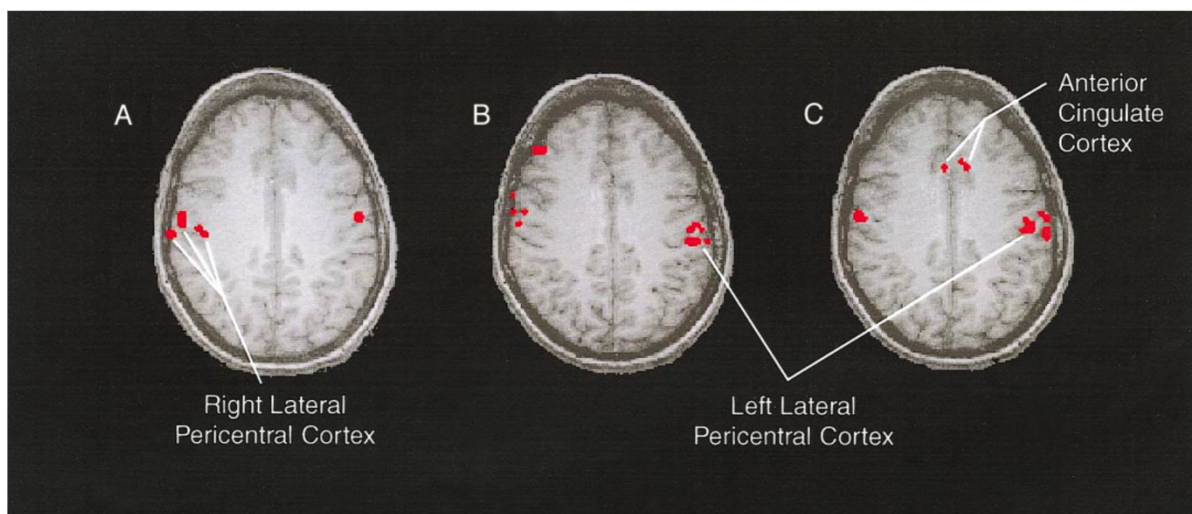


Figure 2.3. Blood-oxygenation-level – dependent (BOLD) activation maps measured in one participant using fMRI during A. naïve saliva swallowing, B. voluntary saliva swallowing, and C. water swallowing. From “Cerebral cortical representation of automatic and volitional swallowing in humans,” by R. Martin, B. Goodyear, J. Gati, & R. Menon, *Journal of Neurophysiology*, Vol. 85, p. 944. Copyright 2001 The American Physiological Society. Reprinted with permission.

Not only have critical regions for neural control of swallowing been identified, the nature of their roles and functions have been postulated (Mosier & Bereznaya, 2001). An fMRI study with healthy adults found that activation of supramedullary structures could be grouped into five functional clusters or modules: 1) the sensorimotor areas (M1, S1, supplementary motor area) and cingulate gyrus, which are thought to be important for the planning, selection, and execution of a sensorimotor sequence, 2) the premotor and parietal cortex, which may be involved in integrating sensory information to plan the motor sequence, 3) the inferior frontal gyrus, secondary sensory cortex, corpus callosum, basal ganglia, and thalamus, which are functionally connected and may be responsible for integrating sensory information about the bolus with internal representation of swallowing movements, 4) the insula, which is believed to play a role in synchronising the timing of movement for bolus control, and 5) the cerebellum, which provides adaptive modulation of oropharyngeal activity to ensure efficient control of movements (Mosier & Bereznaya, 2001). The neural control of swallowing is organised into parallel, functionally-connected cortical areas that mirror the cortical control of other voluntary, skilled behaviours such as writing or speaking (Mosier & Bereznaya, 2001). Damage to these cortical areas important for swallowing may result in higher-level deficits of motor planning, sensory integration, and coordination. For example, lesions to the periventricular white matter, which provides corticocortical connections between sensorimotor areas and is implicated in motor planning, have been associated with lingual discoordination and possible swallowing apraxia in stroke patients (Daniels, Brailey, & Foundas, 1999). Understanding the neural control of swallowing, and the types of swallowing impairments that can arise from neural lesions, is important for management of dysphagia.

The laterality of swallowing cortical control has not been fully established, with studies of laterality demonstrating disparate results across individuals. Hemispheric activation during swallowing has been shown to be bilateral (Mosier, Liu, Maldjian, Shah, & Modi, 1999), lateralized to the left (Martin et al., 2007), or lateralized to the right (Hamdy et al., 1999). Differences in hemispheric activation during swallowing among individuals might be explained by age, with young adults demonstrating right hemisphere dominance while older adults have bilateral representation of swallowing (Malandraki, Sutton, Perlman, & Karampinos, 2010). Type of task might be another factor, with greater sensorimotor cortex activation on the right for volitional swallowing and increased activation in the left hemisphere for reflexive swallowing (Kern, Jaradeh, Arndorfer, & Shaker, 2001). Handedness does not appear to influence laterality (Hamdy et al., 1996). Hamdy et al. (1999) postulated that swallowing representation is greater on one hemisphere than the other, but that the dominant

side differs between individuals, with some having right dominance and others demonstrating left dominance. This individual variance suggests that development of swallowing impairment after unilateral stroke would depend on whether the more dominant swallowing side was damaged, and might explain the large inter-individual differences in severity and duration of impairments (Hamdy et al., 1997).

While neurons in the brainstem provide the basic stereotyped sequence of events during swallowing, the cortex plays a critical role during complex ingestive swallowing involving a bolus. Higher cortical structures are needed to integrate sensory information from the bolus to adapt the motor response in a way that is appropriate and specific for the incoming bolus (Miller, 1999). Adaptations may include modifying the amplitude or duration of the swallowing response depending on cortical and sensory inputs. In this way, cortical control of swallowing allows for a person to modulate their swallowing response to safely ingest boluses of different sizes, consistencies, and temperatures (Daniels & Huckabee, 2014).

In summary, although the basic swallowing pattern generated by the CPG might not be affected by a lesion above the level of the brainstem, knowledge about the cortical control of swallowing suggests that a unilateral cortical lesion can impair motor planning, spatiotemporal precision of motor execution, and the ability to skilfully modulate the swallow response to match environmental needs. This has clinical implications on the assessment and management of patients with swallowing impairments subsequent to neurogenic pathologies.

Chapter 3. Dysphagia

3.1 Definition of dysphagia

Swallowing difficulty, or dysphagia, is abnormal movement of the bolus from the mouth to stomach, and can result from impairment to any of the behavioural, sensory, motor, and/or preparatory acts critical to swallowing (Logemann, 1998; Rosenbek & Jones, 2008). Bolus flow abnormalities are a consequence of impaired kinematics of the structures used for swallowing. For example, in the oral phase, observations of anterior bolus leakage, decreased bolus preparation and formation, or premature spillage to the pharynx are all a result of impaired orolingual control. In the pharyngeal phase, impaired biomechanical movement of the hyoid bone can result in laryngeal penetration or aspiration of the bolus (Logemann, 1998).

Although the relationships between abnormal bolus flow and corresponding biomechanical movement are well understood, the pathophysiology behind impaired biomechanics is less obvious. Deficits in kinematics may be caused by weakness, incoordination, spasticity, or other unexplored mechanisms (Paik et al., 2008). However, the predominant assumption is that biomechanical impairments and dysphagia are caused by underlying weakness. It is important to note that the parameters of abnormal bolus flow and dysfunctional biomechanics can be used to *describe* swallowing impairment, but are not the *cause* of dysphagia. Only underlying pathophysiology can explain why a patient has dysphagia and thus direct the appropriate treatment. There is a critical gap in our knowledge of the dysfunctional mechanisms causing swallowing impairments, which in turn significantly affects the ability to accurately diagnose and treat dysphagia. In the absence of a comprehensive understanding of pathophysiology, and the lack of specific assessments to identify pathophysiologic features, the management of dysphagia has been built on various assumptions.

Many developmental, structural, and neurological aetiologies can cause swallowing impairments (Logemann, 1998). Developmental and congenital disorders causing dysphagia include cerebral palsy and Rett syndrome, affecting feeding and swallowing from birth. Changes in the structures used for swallowing, due to head and neck tumours or the invasive therapies used to treat them, can give rise to dysphagia. Disruption to any aspect of the cortical, brainstem or peripheral levels of swallowing control can cause neurogenic dysphagia, for example in cerebrovascular accident or stroke, Parkinson's Disease, and inflammatory myopathy. For purposes of this thesis, in-depth consideration will be limited to neurogenic dysphagia.

3.2 Pathophysiology of neurogenic dysphagia

There is a “dualism” in the approaches to understanding the underlying mechanisms of dysphagia (Massey & Shaker, 2003, p. 5). One is the traditional swallowing therapy view, which describes dysphagia in terms of disordered biomechanics and impaired bolus flow. Consider, for example, a patient with dysphagia after stroke. The traditional swallowing therapy approach would focus on manoeuvres to change the visualised biomechanical impairments (e.g., supraglottic swallow to facilitate airway closure; Logemann, 1998) and provide the patient with immediate strategies such as postural adjustments and diet changes to cope with dysphagic symptoms. However, this does not provide a long-term solution for eliminating dysphagia or improving swallowing function. The pathophysiologic approach, on the other hand, views swallowing disorders in the context of underlying pathophysiological mechanisms, e.g., weakness or incoordination. Abnormalities of biomechanical movement and bolus flow are the symptoms of such pathophysiological features. This approach targets the underlying disease (e.g., cortical lesion of pyramidal tract resulting in paresis and incoordination) by considering how swallowing has been impacted by the underlying neurological impairment and developing direct rehabilitation strategies to target those pathophysiologic abnormalities. While the visualised biomechanical impairments of two disorders may look similar, they may actually have different underlying causes. Only by targeting the specific underlying pathophysiology can behavioural and neural modifications occur.

Differential diagnosis of the underlying mechanism of dysphagia requires the clinician to consider all possible pathophysiological mechanisms based on knowledge of the lesion location, dysphagic symptoms, and patient history. However, there is an assumption of weakness as the underlying cause of dysphagia, regardless of other factors. It is well known that decreased limb function after neuromuscular lesions can be caused by numerous impairments other than weakness (such as spasticity, impaired motor planning, and sensory deficits), depending on the lesion location along the different levels of motor control (Krakauer, 2005). As indicated in Table 3.1, neurologic disease affects the planning, programming, and execution of limb and motor speech behaviour in such a way that corresponds to the underlying pathophysiology and lesion location (Arene & Hidler, 2009; Duffy, 2005; Raghavan, 2015).

Table 3.1

Pathophysiological Features and Neuromuscular Aetiologies Corresponding to Lesion Location

Location of lesion	Possible pathophysiological features	Examples of neuromuscular aetiologies causing dysphagia
Cortex and brainstem	Weakness Hypertonicity Impaired motor planning Impaired dexterity Impaired sensation	Stroke Traumatic brain injury
Extrapyramidal areas	Weakness Impaired tone and reflexes Hypokinesia Hyperkinesia	Parkinson's disease Huntington's disease
Motor unit	Weakness Hypotonia Impaired sensation	Guillain-Barre syndrome Facial nerve palsy Postpolio syndrome Skull base surgery/tumours Myasthenia gravis
Muscle fibres	Weakness	Muscular dystrophies Polymyositis Dermatomyositis Inclusion body myositis Spinal muscular atrophy

The field of motor speech, which shares many of the structures used in swallowing, acknowledges that neuromuscular lesions can result in patterns of motor speech impairments that can be classified according to site of lesion (Darley, Aronson, & Brown, 1969). Lower motor neuron (LMN) damage causes flaccid dysarthria, while upper motor neuron (UMN) damage causes spastic, unilateral UMN, hypokinetic, hyperkinetic, and ataxic dysarthria. It

would be feasible to hypothesise that a similar neural organisation might be present for swallowing behaviour. There may not be just one type of swallowing impairment (weakness), but many different dysphagia types that systematically mirror the neuroanatomy and neurophysiology of the stroke lesion. Unfortunately, while research has given us broad knowledge regarding the biomechanics and kinematics of disordered swallowing, our knowledge of the underlying mechanisms and aetiologies of dysphagia is lacking in depth.

3.2.1 Cortical and brainstem lesions

The most common aetiology of cortical or brainstem damage resulting in dysphagia is stroke. Stroke patients demonstrate a wide variety of swallowing disorders and symptoms, including increased oral transit time, lingual discoordination, preswallow pooling, delayed pharyngeal swallowing, reduced hyoid elevation, pharyngeal residuals, prolonged pharyngeal transit time, and increased airway invasion (Daniels & Huckabee, 2014; Perlman, Booth, & Grayhack, 1994; Robbins, Levine, Maser, Rosenbek, & Kempster, 1993). Possibly because of the complexity of neural control of swallowing, there is large individual variation in dysphagia symptoms after neurological damage, even between patients with similar lesions. Studies have attempted to correlate dysphagia symptoms by lesion location (Daniels & Foundas, 1999; Steinhagen, Grossmann, Benecke, & Walter, 2009), hemisphere (Daniels et al., 1996; Robbins et al., 1993), or size (Alberts, Horner, Gray, & Brazer, 1992), but results have been inconsistent. Patients with right hemisphere strokes have been found to have more pharyngeal abnormalities, whereas left hemisphere stroke patients have more oral phase problems (Daniels et al., 1996; Robbins et al., 1993). Lesion hemisphere has been found to be a significant predictor of airway invasion risk (Daniels et al., 2017). However, other studies have shown that hemispheric location was more important than right/left hemisphere in predicting risk of aspiration (Daniels & Foundas, 1999).

Other studies have attempted to find a relationship between swallowing impairment pattern and lesion size. Daniels & Foundas (1999) found that size of lesion was less important than location in predicting patients at risk of aspiration, with most of the patients with dysphagia in the study having small or medium-sized lesions. However, another study concluded that size, rather than location, was more associated with swallowing dysfunction (Paciaroni et al., 2004). One reason for the inconsistent correlations between lesion site and dysphagia symptoms could be the asymmetrical representation of swallowing in the cerebral cortex (Hamdy et al., 1996). There is a dominant swallowing hemisphere (right or left) that differs between individuals, and

dysphagia may be more likely if the stroke occurs in the patient's dominant versus non-dominant hemisphere (Hamdy et al., 1997).

Examination of the limb and motor speech literature can provide an indication as to the types of impairment expected after central neural damage. UMN lesions often affect both the direct activation pathway, which controls skilled and precise movements, as well as the indirect activation pathway, which regulates reflexes and muscle tone (Duffy, 2005). Therefore, lesions at this level result mainly in weakness, hypertonia and hyperreflexia, impaired motor planning, and loss of skilled motor execution (Duffy, 2005; Raghavan, 2007).

3.2.1.1 Weakness

Weakness is defined as an impairment in producing adequate muscular force or strength (Ng & Shepherd, 2000), but it can also manifest as decreased speed to generate force (Canning, Ada, & O'Dwyer, 1999), increased sense of effort (Patten, Lexell, & Brown, 2004), and increased sense of fatigue (Vøllestad, 1997). Adequate muscular force depends on both muscular factors (i.e., cross-sectional area of the muscle) and neural factors (i.e., ability of the nervous system to recruit motor units and activate muscle; Patten et al., 2004). Compromise to either or both factors results in the reduced capacity to generate force. Weakness in the limbs after stroke and other UMN lesions is predominantly caused by direct neural changes to agonist motor units, with decreased number of functioning motor units and reduced motor unit firing rates (Arene & Hidler, 2009; Ng & Shepherd, 2000). There are also secondary adaptive changes in the muscles, such as muscle fibre shortening (contracture) and atrophy due to immobility and misuse over time (Ng & Shepherd, 2000). In addition to these two mechanisms of weakness, there may also be indirect processes that restrict agonist activation, for example spasticity (velocity-dependent hyperactive stretch reflexes) in the antagonist muscle, changes in mechanical properties of the antagonist muscle, and excessive and inappropriate cocontraction of the agonist and antagonist muscle groups (Bourbonnais & Vanden Noven, 1989; Gracies, 2005b; Ng & Shepherd, 2000). However, current research demonstrates that spasticity does not play a major role in limiting the production of muscular force in the limbs (Bohannon, Larkin, Smith, & Horton, 1987; Bourbonnais & Vanden Noven, 1989; Gracies, 2005b; Ng & Shepherd, 2000).

In the motor speech system, unilateral UMN damage causes relatively mild effects on the muscles of the jaw, pharynx, soft palate, and larynx. Innervation of the cranial nerves for muscles of motor speech is bilateral, with the exception of the predominantly contralateral

innervation of the lower face and tongue. Since the direct and indirect activation pathways have direct input to the LMNs, weakness can still occur after a UMN lesion, but to a lesser extent compared to LMN lesions (Duffy, 2005).

It is commonly assumed that swallowing impairment after a stroke is a result of decreased strength, with the majority of dysphagia rehabilitation techniques targeted toward increasing strength (Rogus-Pulia & Robbins, 2013). However, little is known about the mechanisms of weakness in the oropharyngeal musculature after a stroke, and how this weakness affects functional swallowing. While patients with dysphagia may have accompanying muscular weakness, the degree to which weakness causes dysphagia and influences functional swallowing remains unclear (Clark, 2003; Clark, Henson, Barber, Stierwalt, & Sherrill, 2003).

3.2.1.2 Hypertonicity

Muscle tone is defined as the resistance provided by the muscle to external movement. When a muscle is passively stretched, muscle spindles are activated. The muscle spindles transmit impulses directly onto lower motor neurons at the brainstem or spinal cord, which in turn causes the muscle to contract and resist the stretch. This peripheral stretch reflex is further regulated and inhibited by descending cortical and subcortical pathways (Clark & Solomon, 2012b; Duffy, 2005; Shumway-Cook & Woollacott, 2017). Hypertonicity in the limbs manifests as spasticity and increased muscle stretch reflexes. Spasticity is defined as increased velocity-dependent stretch reflex activity in passive movement (Gracies, 2005b). While spasticity in the limbs can develop and contribute to disability after stroke or traumatic brain injury, the traditional view that spasticity is the main detriment to motor functioning has not been supported by more recent research (Bohannon et al., 1987; Patten et al., 2004; Sommerfeld, Eek, Svensson, Holmqvist, & von Arbin, 2004).

Despite the lack of research on spasticity in the swallowing mechanism, the effect of spasticity on motor speech has been documented (Darley et al., 1969; Duffy, 2005). It is reasonable to surmise that if UMN damage can cause spasticity in motor speech, there is potential for spasticity to affect other corticobulbar functions such as swallowing (Huckabee & Kelly, 2006). Spasticity in the laryngeal musculature is associated with vocal hyperadduction, resulting in the characteristic strained-strangled vocal quality present in spastic dysarthria (Duffy, 2005). Patients with spasticity (e.g., from cerebral palsy) often have oral and pharyngeal swallowing disorders, as assessed with videofluoroscopy, ultrasound, and clinical swallowing examination (van den Engel-Hoek et al., 2014; Vogel, Brown, Folker, Corben, &

Delatycki, 2014). However, when comparing the swallowing disorders of two groups of children with cerebral palsy, researchers found that the group with spastic features did not differ significantly from the group with dyskinetic features, making it difficult to elucidate the true contribution of spasticity to dysphagia (van den Engel-Hoek et al., 2014).

Spasticity and weakness in the limbs often occur together after stroke because of the proximity of the direct and indirect activation pathways (Krakauer, 2005; Raghavan, 2015), and as discussed previously, spasticity may contribute to weakness. Thus, it is important to consider the possibility of abnormal muscle tone underlying dysphagia in stroke patients. In addition, since both hypertonicity and weakness in the limbs can manifest as reduced range of motion, patients with spasticity may be misdiagnosed with poor strength and vice versa, resulting in ineffective treatment strategies (Bourbonnais & Vanden Noven, 1989). A lack of specific and objective tests to measure resistance to passive stretch (i.e., hypertonicity) in the oropharyngeal musculature has compounded this problem. Current clinical assessment of muscle tone is limited to perceptual rating scales of stretch and palpation (Clark & Solomon, 2012b). A few exploratory studies have investigated the use of instrumental assessments for measuring oropharyngeal tone, including an accelerometer that measures tongue stiffness from a brief pulse perturbation (Dietsch et al., 2014; Solomon & Clark, 2010) and pharyngeal sEMG to measure changes in muscle activity (Doeltgen et al., 2007). However, initial results show that these methods were not sensitive to detecting changes in different conditions, resulted in high inter- and intra-participant variability, and had methodological issues which impacted the validity of the measure.

3.2.1.3 Impaired motor planning

In the limb and motor speech literature, damage to the motor planning and programming levels is unlikely to cause significant muscular weakness. These levels of control, as represented by premotor and supplementary motor areas in the cerebral cortex, do not have direct influence on the cranial nerves and muscles. Instead, apraxia is a common finding, defined as the impairment in motor planning and programming of a skilled action, not due to any other impairment in motor, sensory, or language functioning (Gross & Grossman, 2008; Koski, Iacoboni, & Mazziotta, 2002). This is different from impaired dexterity, which is an impairment in motor execution. In apraxia, the meaningful, learned skilled behaviour can often be executed automatically in the context of everyday situations, but the patient is unable to do so voluntarily or by imitation. Since motor planning and programming are high-level processes controlled at the level of the cortex, apraxic speech and limb movement most commonly result

from focal lesions to the left cerebral hemisphere, for example after stroke, tumours or surgical trauma (Duffy, 2005).

Many types of apraxia have been identified in the context of voluntary behaviours, such as limb apraxia (Gross & Grossman, 2008), orofacial apraxia (Ozsancak, Auzou, Dujardin, Quinn, & Destée, 2004), and apraxia of speech (Duffy, 2005). Apraxia of swallowing has been suggested in the literature as a motor planning impairment in the oral phase of swallowing, separate from motor execution deficits. The definition of swallowing apraxia, and the idea that it can be classified as a true type of apraxia, has been under debate (Daniels, 2000). While there has been limited research completed on swallowing apraxia, it has generally been described as random, disorganised oral movements resulting in difficulty with anterior-to-posterior bolus propulsion during the oral phase, despite normal range of motion (Logemann, 1998). Similar to the other apraxias, swallowing apraxia is characterised by reduced performance upon command, improved movement in natural environments, difficulty with initiation of movement, and spatiotemporal errors (Daniels et al., 1999, 1996; Robbins et al., 1993). Swallowing apraxia also shares a common site of lesion with limb apraxias, as they are associated with damage to the cortical left hemisphere (Robbins et al., 1993). However, another study found that swallowing apraxia was not predicted by right or left lesion hemisphere (Daniels et al., 1999). There are key differences between swallowing and other behaviours that have raised questions about whether swallowing apraxia can be classified as a disorder of the praxis system. Definitions of apraxia emphasise a disorder of “learned” movement (Geschwind, 1975), while swallowing is a patterned response that is not learned. While other forms of apraxia can be evaluated using multiple gestures (e.g., limb gesture to command, gesture to imitation, gesture with and without an object; Dovern, Fink, & Weiss, 2012), swallowing apraxia can be assessed with only a single gesture, that is, swallowing. The incoordination could be due to upper motor neuron or sensory impairments, instead of deficits in motor planning. Spatial and temporal errors have been noted in all the other apraxias, but have not been studied in swallowing apraxia. In order to fully define swallowing apraxia, specific temporal and spatial error patterns need to be identified, and more specific assessment tools need to be used to discriminate incoordination due to motor planning and incoordination due to other impairments (Daniels, 2000).

3.2.1.4 Impaired skill

Along with decreased strength, loss of motor skill in the limbs is the main negative feature of UMN lesions to the direct activation pathway (Canning, Ada, & O’Dwyer, 2000). A related

concept is that of impaired dexterity, which has been defined as the impaired coordination of skilled, voluntary movements to meet environmental demands. Decreased skill and dexterity have been found to be an independent and significant factor contributing to impaired function after stroke (Canning, Ada, Adams, & O'Dwyer, 2004; Canning et al., 2000). Impaired dexterity is characterised by reduced spatial and temporal precision of limb movement, usually assessed during performance of a visuomotor tracking task (Canning et al., 2000; Van Hedel, Wirth, & Curt, 2010). It is unknown whether the definition of impaired dexterity in voluntary movement can be applied to “innate”, central-pattern-generated behaviours such as respiration, mastication and swallowing. While the CPG is responsible for the basic patterned swallowing response, cortical processes provide substantial and volitional modulation of this basic pattern to respond to environmental demands. Given that swallowing is a complex, coordinated sequence of movements, it stands to reason that impaired spatial and temporal movement precision may be a factor contributing to dysphagia.

While the concept of movement skill or dexterity in swallowing has gone relatively unexplored in the literature, there is some preliminary evidence of impaired coordination contributing to dysphagia. The use of temporally-sensitive assessments has demonstrated dyscoordinated sequencing of pharyngeal pressure generation in patients with dysphagia from infratentorial lesions (Figure 3.1; Huckabee, Lamvik, & Jones, 2014), as well as abnormal timing and patterning of respiratory-swallowing events in Parkinson's patients (Troche, Huebner, Rosenbek, Okun, & Sapienza, 2011). In addition, impaired motor control in a brainstem stroke patient with dysphagia was evidenced by their decreased accuracy in controlling amplitude and timing of pharyngeal and laryngeal muscle activity to achieve an on-screen target using visual feedback, compared to healthy controls (Stepp, Britton, Chang, Merati, & Matsuoka, 2011). However, no research has been completed measuring the relative contribution of skill impairment, independent of weakness, in patients with supratentorial stroke.

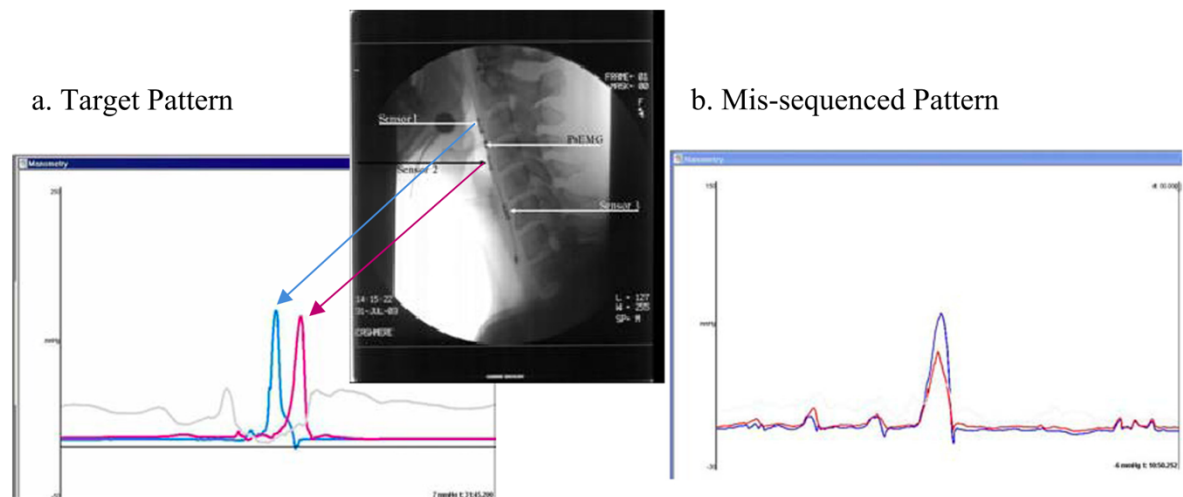


Figure 3.1. Pharyngeal manometry waveforms. Panel a demonstrates a normal pattern of pharyngeal pressure generation, where peak-to-peak latency between nadir pressures at the proximal (blue waveform) and distal (red waveform) pharynx was greater than 200 ms. Panel b demonstrates an abnormal, mis-sequenced pattern where the pressure peaks are generated simultaneously. From “Pharyngeal mis-sequencing in dysphagia: Characteristics, rehabilitative response, and etiological speculation,” by M. Huckabee, K. Lamvik, & R. Jones, *Journal of the Neurological Sciences*, Vol. 343, p. 156. Copyright 2014 by Elsevier. Reprinted with permission.

3.2.1.5 Impaired sensation

In the limbs, sensory impairment can occur with central (e.g., parietal lobe) and/or peripheral lesions, and it affects the degree to which patients can participate in activities of daily living as well as recovery time (Raghavan, 2015). In swallowing, deficits in sensation can result in the inability of the central nervous system to integrate information about the incoming bolus and modulate the motor output accordingly. Patients with brainstem and supratentorial stroke were found to have significant sensory impairments compared to healthy controls, as measured by patients having greater sensory discrimination thresholds for air pulses delivered to the pharyngeal mucosa via flexible fiberoptic telescope (Aviv et al., 1996). However, there was no clear relationship between the sensory deficit and the severity of dysphagia assessed clinically. Sensory impairments have been associated with functional swallowing problems, including delayed initiation of pharyngeal swallowing (Logemann, 1998; Martin-Harris et al., 2007), impaired airway protection (Jafari, Prince, Kim, & Paydarfar, 2003), and aspiration (Setzen et al., 2003). However, the role and mechanism of sensory impairment in relation to swallowing has not been well-studied in the dysphagia literature. Oral and pharyngeal sensation are not

routinely assessed in instrumental and clinical evaluation of swallowing, even though it is recognised that sensory input plays an important part in the initiation and modulation of the swallowing response (Steele & Miller, 2010).

3.2.2 *Extrapyramidal disorders*

Damage localised to the basal ganglia is unlikely to cause significant muscular weakness in the limbs and for motor speech, since this subcortical area does not have direct influence on the cranial nerves and muscles. The basal ganglia maintains muscle tone, posture, and static muscle contraction, and also has an inhibitory and modulatory influence on cortical pathways (Duffy, 2005). Basal ganglia lesions result in movement disorders, namely hyperkinesia and hypokinesia.

3.2.2.1 Hypo- and hyperkinesia

Hypokinetic disorders are related to the pathological increase in basal ganglia output with greater inhibition of voluntary movement, resulting in the characteristic slow and stiff limb movements, reduced range of motion, and masked faces seen in Parkinson's Disease (Wichmann & DeLong, 1996). Hyperkinetic disorders such as Huntington's Disease are associated with decreased basal ganglia output and disinhibition of cortical impulses, resulting in involuntary, excessive and unpredictable limb movements (Wichmann & DeLong, 1996). Since these movement disorders are observed in the limbs, they are also thought to be the major contributors to the deficits seen in oral and pharyngeal swallowing. For example, the assumption is that bradykinesia and rigidity underlie swallowing deficits in Parkinson's Disease, which are characterised by lingual pumping, decreased bolus formation and control, reduced initiation of pharyngeal swallowing, and pharyngeal dysmotility (Robbins, Logemann, & Kirshner, 1986). However, bradykinesia and rigidity in the swallowing musculature have not been assessed directly.

3.2.3 *Motor unit abnormalities*

A motor unit is made up of the lower motor neuron and the muscle fibres it innervates (Duffy, 2005; Merletti & Parker, 2004). Damage to the lower motor neuron (LMN) is characterised by variable levels of weakness, reduced sensation, and reduced muscle tone in the limbs and motor speech mechanism, resulting in decreased rate, range of motion, and accuracy of movements (Duffy, 2005). Since each muscle is innervated by several motor neurons, damage to a single motor neuron means that muscle contraction can still occur. If the LMN is only partially

damaged and there is some remaining input to the muscle fibres, this results in paresis, or reduced contraction. If all innervation to the muscle is severed from motor neuron activation, then this results in paralysis, or the complete inability of muscles to contract. Common causes of partial or complete LMN damage include postpolio syndrome (Sonies & Dalakas, 1991), surgery or tumours in the skull base or cranial nerves (Peterson & Fenn, 2005), Guillain-Barré syndrome, and facial nerve palsy. In swallowing, unilateral damage to the LMNs (i.e., cranial nerves) results in paresis (usually ipsilateral) of the oropharyngeal muscles (Périé et al., 1999).

Damage at the neuromuscular junction (such as in myasthenia gravis) can also result in weakness and fatigue. Neuromuscular junction insult results in decreased numbers of functioning acetylcholine receptors at the postsynaptic membrane. The reduced availability of acetylcholine results in progressively decreasing muscle contraction with repeated use, but muscle strength can recover with rest as acetylcholine reserves are replaced (Duffy, 2005).

3.2.3.1 Hypotonia

LMN damage can result in muscle tone impairments, but impairments are different from those seen in UMN damage. Hypotonia, or reduced muscle tone, can be caused by LMN damage because the muscle is unable to send efferent impulses to the muscle to contract and resist the stretch reflex (Duffy, 2005). Hypotonia is seen in flaccid and ataxic dysarthria (Duffy, 2005). In the limb literature, hypotonicity is different from weakness: hypotonicity is poor resistance to passive movement, whereas muscle weakness is the reduced ability to generate active movement. In addition, a patient can present with hypotonia and still have intact strength (Leyenaar, Camfield, & Camfield, 2005), highlighting that the impairments are independent of each other and can have different effects on functioning. Hypotonia in the swallowing system is not measured as resistance to passive movement, but instead assessed using lingual pressure and subjective ratings of oral-motor sensation (Hashimoto et al., 2014; Siktberg & Bantz, 1999). Hypotonia in the oral musculature is thought to affect the duration and magnitude of tongue movement and impair oral sensation, resulting in oral phase dysphagia (Hashimoto et al., 2014; Miller & Britton, 2011; Siktberg & Bantz, 1999). However, the lack of diagnostic specificity means that the definition and assessment of hypotonia in swallowing is likely confounded by muscle weakness and/or sensory deficits.

3.2.4 Myopathy

Weakness is the most significant characteristic associated with myopathy, which is defined as pathology and atrophy at the level of skeletal muscle (Miller & Britton, 2011). The main muscular factor determining force generation is the cross-sectional area of the muscle itself (Burkhead, Sapienza, & Rosenbek, 2007; Merletti & Parker, 2004). Therefore, muscle atrophy and fibre necrosis seen in muscular disorders can result in substantial weakness (Briani, Doria, Sarzi-Puttini, & Dalakas, 2006; Turner & Hilton-Jones, 2010). Damaged muscle fibres can be caused by several mechanisms, including infiltration of inflammatory cells in the muscle tissue (inflammatory myopathy; Loell & Lundberg, 2011) and genetic mutations of muscle cell proteins (muscular dystrophy; Goldstein & McNally, 2010).

Inflammatory myopathies often present with dysphagia in all phases of swallowing. Of the four inflammatory myopathy types, dysphagia is most severe in inclusion body myositis (IBM), with aspiration pneumonia being the main cause of death in this population (Langdon, Mulcahy, Shepherd, Low, & Mastaglia, 2012; Peng, Koffman, Malley, & Dalakas, 2000). Dysphagia is also prevalent in two types of muscular dystrophy: myotonic muscular dystrophy (myotonic MD) and oculopharyngeal muscular dystrophy (OPMD; Miller & Britton, 2011). In these muscular disorders, weakness is the primary mechanism of dysphagia, as evidenced by significantly reduced peak pharyngeal pressure measured with manometry (Langdon et al., 2012), increased pharyngeal constriction ratios on VFSS (Leonard, Kendall, Johnson, & McKenzie, 2001), and reduced isometric and swallowing lingual pressure, compared to age-matched healthy controls (Palmer, Neel, & Morrison, 2010). This underlying weakness likely causes impaired swallowing function, as patients with OPMD have decreased swallowing capacity and volume per swallow during a water swallowing test. Decreased maximum isometric intraoral pressure has been shown in myopathic patients to be significantly correlated with reductions in swallowing pressure, swallowing capacity, and swallowing-related quality of life (Palmer et al., 2010).

3.2.5 Summary

In summary, a range of sensorimotor impairments can occur after neuromuscular damage to the central or peripheral nervous system, including but not limited to weakness, impaired tone, sensory deficits, and reduced coordination. For patients with damage at the level of the muscle, as in myopathic diseases, weakness would be considered the cardinal feature. However, for central lesions such as supratentorial stroke, decreased dexterity and precision of movement, increased tone, and weakness may be expected instead. Both upper and lower motor neuron lesions can result in weakness, but the mechanisms causing weakness are different in central

versus peripheral damage. The same biomechanical impairments can be caused by a variety of pathologies (e.g., reduced range of motion can be visualised in patients with hypertonicity, weakness, and/or impaired dexterity). Therefore, the different pathologies underlying dysphagia can be confused with weakness. The ability to differentiate weakness from other impairments would be critical for improved patient care. Even though neurological disorders may be characterised by the presence of certain pathophysiological features in the limb or speech musculature, this does not necessarily translate to the swallowing mechanism. This assumption requires specific testing before acceptance. The assumption of weakness as the main cause of dysphagia is likely due to the lack of specific and objective assessments which can differentiate between the underlying mechanisms of dysphagia.

3.3 Consequences of dysphagia

Rosenbek & Jones (2008) emphasize that normal swallowing must meet all of the following conditions: *safe*, *efficient*, and *satisfying*. When swallowing is impaired, the consequences can be serious and can affect the safety, efficiency, and satisfaction of swallowing to varying degrees. Swallowing impairments can endanger the safety of the patient by increasing the risk of chest infection and malnutrition, resulting in inefficient and laborious meal times, that negatively impact quality of life (Martino et al., 2005). Depending on how dysphagia is identified, the incidence of stroke patients who develop dysphagia ranges from 37 to 78% (Martino et al., 2005). Mann, Hankey, & Cameron (1999) assessed swallowing in first-time stroke patients and found that 51% of them had clinically-detected dysphagia, while dysphagia was detected videofluoroscopically in 64% of the patients. After six months, swallowing evaluations were completed again on those with dysphagia. It was found that 50% continued to have persistent dysphagia diagnosed clinically, while videofluoroscopy detected dysphagia in 81% of the patients, and 13% had not returned to their pre-stroke diet. Therefore, swallowing impairments can persist past the acute stage, with negative impact on functional swallowing outcomes.

Aspiration pneumonia is a serious and life-threatening consequence of dysphagia, leading to hospitalisations and re-admissions, prolonged length of stay, increased healthcare costs, and mortality (Baine, Yu, & Summe, 2001). It occurs when oropharyngeal material (food, liquid, or secretions) are colonised by pathogens and aspirated into the lungs. If the material is unable to be cleared and becomes infected, the person with dysphagia can develop aspiration pneumonia (Langmore et al., 1998; Marik & Kaplan, 2003). A systematic review of dysphagia and aspiration in stroke found that of the acute stroke patients referred for an instrumental

swallowing exam, 19.5% to 42% aspirated (Perry & Love, 2001). An even larger proportion (53.5%) of stroke patients in a rehabilitation setting demonstrate aspiration; 39% of them aspirate silently, without coughing or attempts at clearing the airway (Holas, DePippo, & Reding, 1994). While the presence of dysphagia and aspiration alone is not enough to cause aspiration pneumonia, dysphagia and aspiration remain important risk factors for developing pneumonia (Langmore et al., 1998). The relative risk of pneumonia is higher (3.17) for patients with dysphagia than those without dysphagia, and even higher (11.56) for patients who aspirate compared with those who do not aspirate (Martino et al., 2005). In addition to aspiration pneumonia, aspiration of the bolus into the respiratory tract can cause airway obstruction. Patients with dysphagia are more likely to have choking episodes (Ekberg & Feinberg, 1992). Compensatory strategies such as feeding tubes and diet modifications are not effective at preventing aspiration or pneumonia. Further understanding of the physiological mechanisms of dysphagia and aspiration in stroke patients can lead to better prevention and treatment of aspiration pneumonia.

Patients who have post-stroke dysphagia have an increased risk of malnutrition, with a higher odds ratio of being malnourished compared to those without dysphagia. It is also common for patients with dysphagia to have inadequate fluid intake and dehydration (Leibovitz et al., 2007). This may be for several reasons – disordered swallowing biomechanics (e.g., reduced UES opening) may result in insufficient amounts of the bolus entering the oesophagus and stomach. In addition, fear of choking, decreased palatability of prescribed modified diets, and increased feelings of satiety when drinking thickened liquids can result in inadequate intake (Cichero, 2013; Foley, Martin, Salter, & Teasell, 2009). Over half of people with dysphagia report eating less because of discomfort and difficulty during swallowing, resulting in lethargy and weight loss (Ekberg, Hamdy, Woisard, Wuttge-Hannig, & Ortega, 2002).

Eating and drinking are not only for obtaining adequate nutrition and hydration, but are also social and enjoyable experiences. Food and drink are an integral part of family gatherings and celebrations, and provide an opportunity for people to connect with others in their community. Missing out on these social experiences can cause feelings of isolation, decreased self-worth, anxiety and sadness, which drastically reduces overall quality of life (Ekberg et al., 2002). Given the serious health, nutritional, and psychological consequences of dysphagia, it is imperative that accurate diagnosis of impairments is provided to patients in a timely manner.

3.4 Assessment of dysphagia

Differential diagnosis of the specific mechanism underlying a patient's dysphagia should include the careful consideration of all the possible pathophysiological features that can result, based on neuroanatomical location of the lesion (Heneghan et al., 2009). For example, as discussed in a previous section, a stroke patient might exhibit weakness, impaired dexterity, and other deficits. The patient's symptoms, history, and results from clinical and instrumental tests can then be used to rule in or rule out these possible pathophysiological features. However, current assessment methods are lacking in the ability to identify or discriminate between the different mechanisms underlying dysphagia. Providing treatment without knowing the true underlying problem is a waste of resources and may even exacerbate the patient's dysphagia (Garcia, Hakel, & Lazarus, 2004). Misdiagnosis of swallowing disorders may result in increased risk for aspiration pneumonia, inappropriate diet recommendations, and development of ineffective rehabilitation strategies (Pikus et al., 2003; Splaingard, Hutchins, Sulton, & Chaudhuri, 1988).

3.4.1 Clinical examination

The clinical swallowing examination typically involves a review of the patient's history, cranial nerve assessment, and observation of behaviours during oral intake of liquids and/or solid food. Given the serious consequences of dysphagia, it is imperative that there is accurate and timely identification of dysphagia in the acute setting to determine whether a patient requires further instrumental evaluation. Early screening and assessment of dysphagia is associated with reduced pneumonia rates, decreased mortality, reduced length of hospital stay, and increased cost effectiveness (Bray et al., 2017; Martino, Pron, & Diamant, 2000; Odderson, Keaton, & McKenna, 1995; Odderson & McKenna, 1993; Titsworth et al., 2013). There are other advantages to the clinical assessment: it is inexpensive, it does not require specialised equipment, and can be conducted in any setting, including at the patient's bedside. However, there is substantial variability between speech-language pathologists (SLPs) in the choice of oral motor tasks and bolus trials used in clinical assessment (McCullough, Wertz, Rosenbek, & Dinneen, 1999), as well as variability in how decisions are made based on the clinical exam results (Mathers-Schmidt & Kurlinski, 2003). In one study, SLPs were given six patient case scenarios containing clinical swallowing assessment results, and asked to recommend either a follow-up instrumental assessment, non-instrumental assessment, or other course of action (Mathers-Schmidt & Kurlinski, 2003). There was agreement (over 80%) between the clinicians in only two of the six patient scenarios. Disagreement on recommendations could not be explained by clinicians' experience working with dysphagia or availability of instrumentation. These inconsistencies in SLP practice are likely due to the lack of research on clinical

assessment measures, and highlights the need for the development of measures that are standardised, valid and reliable (McCullough et al., 1999).

Due to the subjective nature of the clinical swallowing examination, research has questioned the cost-effectiveness and reliability of these protocols (McCullough et al., 2000; Wilson & Howe, 2012). Clinicians often evaluate strength of muscles involved in swallowing by assessing the ability of oral structures to push against or overcome some level of resistance provided by the clinician (e.g., jaw opening against the clinician's hand). Coordination may be evaluated by assessing the speed and regularity of alternating movements (e.g., coordination of side-to-side lingual movement). However, there are no standards regarding how much resistance a clinician should provide, or how to judge whether coordination is within normal limits (Clark, 2005). During clinical exam, it is impossible to visualise the bolus and internal structures during swallowing. In addition, many swallowing structures in the pharynx and larynx are not visible to assess coordination at bedside, nor are they accessible to assess resistance. It is no surprise, then, that coordination and impairments other than weakness have not formed the basis of any assessment or treatment approaches. This also explains why clinicians may be forced to make judgements of swallowing physiology based on subjective inferences and qualitative speculations. Less than half of clinical measures were rated with sufficient inter- and intrajudge reliability (McCullough et al., 2000). Further, these clinical ratings are usually made in a binary manner (normal or abnormal), making it difficult to quantify severity of impairment and degree of change over time.

Clinicians also use signs and symptoms observed during a clinical exam (e.g., coughing or throat clearing during trials of oral intake) to predict possible aspiration. However, the clinical detection of silent aspiration (that is, entry of food and liquid past the vocal folds and into the lungs, without a cough response) is particularly challenging. Splaingard, Hutchins, Sulton, & Chaudhuri (1988) found that clinical examination was unable to identify 58% of aspirating patients, and missed 70% of patients who were profoundly aspirating on videofluoroscopy. Attempts have been made to increase the objectivity and accuracy of the clinical examination with adjunct instrumentation. Cough reflex testing has been documented as a promising tool for identifying patients who silently aspirate (Miles et al., 2013). The test uses a nebulised tussive agent (e.g., citric acid or capsaicin) inhaled via facemask, which stimulates laryngeal sensory receptors and induces a reflexive cough response in a healthy system. However, patients with neurological injury may have a weak or absent cough response to the test, indicating they have lost this protective mechanism, increasing their risk of aspiration

pneumonia (Widdicombe, Addington, Fontana, & Stephens, 2011). Incorporating results of cough reflex testing into the clinical swallowing evaluation is associated with better patient outcomes, including decreased pneumonia rates, shorter hospital length of stay, and improved diet level (Addington, Stephens, Gilliland, & Rodriguez, 1999; Davies, 2016). Pulse oximetry and cervical auscultation have been proposed as simple and low-cost adjuncts to the clinical examination, but their clinical utility remains widely contested due to inadequate reliability and validity in identifying aspiration (Higo, Tayama, Watanabe, & Nito, 2003; Lagarde, Kamalski, & van den Engel-Hoek, 2015; Wang, Chang, Chen, & Hsiao, 2005).

It is important to note that the overwhelming majority of research focuses on the ability of the clinical exam to predict the presence or absence of aspiration, but not every patient with dysphagia demonstrates aspiration (Daniels, McAdam, Brailey, & Foundas, 1997). Focusing solely on the presence of aspiration can result in patients with significant dysphagia unidentified, leaving them at high risk for malnutrition and dehydration, and reduced quality of life. In addition, important information that contributes to a better understanding of a patient's overall swallowing physiology and potential for rehabilitation may be missed (Daniels & Huckabee, 2014). One study investigated the ability of clinical assessment to accurately predict dysphagia, not just aspiration, as identified on VFSS (Mann & Hankey, 2001). Significant clinical predictors of dysphagia were pharyngeal response impairment, incomplete oral clearance, palatal asymmetry, stroke severity, male gender, and age over 70 years. However, their definition of dysphagia on VFSS was based on abnormal bolus flow, and not physiological measures of swallowing. This underscores the need for research on clinical measures that can identify patients at risk of dysphagia and the underlying pathophysiology of dysphagia, not just aspiration.

Quantitative measures of oral intake, such as timed water swallowing tests and the Test of Masticating and Swallowing Solids (TOMASS), may provide more objective and reliable information about swallowing function than traditional pass/fail screenings (Athukorala, Jones, Sella, & Huckabee, 2014; Hughes & Wiles, 1996; Nathadwarawala, Nicklin, & Wiles, 1992). In these assessments, patients ingest a standardised liquid or solid bolus as quickly as is comfortably possible, while the clinician takes measurements such as number of swallows and total time taken. Both of these quantitative swallowing tests have been shown to have high inter- and intra-rater and test-retest reliability (Huckabee et al., 2018; Nathadwarawala et al., 1992), which make them valuable for monitoring change in swallowing function over time. Normative values have been established for both males and females in different age groups,

which can improve diagnosis and classification of normal versus abnormal swallowing (Huckabee et al., 2018; Hughes & Wiles, 1996; Nathadwarawala et al., 1992). However, these tests have not been fully validated against instrumental assessments, and do not provide information on the nature or severity of underlying pathology or impairment (Hughes & Wiles, 1996).

In addition to clinician-observed measures, it is important to document quality of life and patient-reported outcomes to address the social, emotional and psychological consequences of dysphagia. The EAT-10 is a 10-item questionnaire which uses a 5-point scale to rate symptom severity (0 = no problem; 4 = severe problem), with a total score ranging between 0 and 40 (Belafsky et al., 2008). Normative data suggest that a total score of 3 or more is indicative of abnormal swallowing. Research has shown that the EAT-10 has good validity and reliability, as well as high sensitivity and specificity for detecting oropharyngeal dysphagia, but poor specificity for predicting aspiration (Cheney, Siddiqui, Litts, Kuhn, & Belafsky, 2014; Rofes, Arreola, Mukherjee, & Clavé, 2014). Although quality of life questionnaires are an essential adjunct to the swallowing evaluation, it does not provide information on swallowing biomechanics or pathophysiology.

3.4.2 Instrumental examination

Instrumental swallowing examinations allow for more objective measurement of swallowing function, but as discussed below, each assessment in isolation may not provide the comprehensive information needed to understand the complex mechanisms underlying dysphagia. These assessments should be seen as complementary, as each has strengths and weaknesses in its ability to evaluate different aspects of swallowing. The most commonly used methods in research and clinical practice are videofluoroscopic swallowing studies (VFSS), fiberoptic endoscopic evaluation of swallowing (FEES), and pharyngeal manometry.

VFSS is often considered the “gold standard” of swallowing assessments as it offers a comprehensive view of the oral, pharyngeal, and oesophageal phases of swallowing as an integrated process. It provides dynamic and real-time videoradiographic images of swallowing biomechanics and bolus flow as different food and liquid consistencies (impregnated with barium) travel from the oral cavity to the upper oesophagus (Logemann, 1998; Martin-Harris & Jones, 2008). Based on these images, judgements are then made regarding the abnormal physiology that may be causing the swallowing impairment. For example, reduced anterior hyoid movement and incomplete epiglottic inversion may be seen, resulting in redirection of

the bolus into the open airway. While one might assume weakness as a cause for these biomechanical and bolus flow abnormalities, it is imperative to understand that VFSS does not directly measure the presence or severity of sensory and motor impairments (Martin-Harris & Jones, 2008). The biomechanical abnormality of reduced hyoid movement is often assumed to be caused by the underlying pathophysiology of muscle weakness. However, as noted in previous sections, there are other neuromuscular abnormalities besides weakness that can result in impaired biomechanical movement. Visualisation of reduced hyoid bone movement on VFSS may suggest weakness but does not rule out the presence of other causes, such as impaired movement precision or coordination. VFSS lacks the diagnostic specificity to directly assess underlying pathophysiology of biomechanical deficits, which in turn affects our ability to apply specific treatments (Huckabee & Macrae, 2014).

Besides this lack of diagnostic specificity, there are other disadvantages to VFSS, including radiation exposure, which limits the duration of an examination, the need for radiology equipment and staff, high cost, and inability to be used at bedside. The reliability and measurement accuracy of VFSS measurements have also been questioned. Different swallowing parameters may be judged by the clinician on a binary yes/no basis (Perlman, Grayhack, & Booth, 1992a) or using descriptive rating scales (Stoeckli, Huisman, Seifert, & Martin-Harris, 2003). Inter-rater reliability has been found to be low, with kappa coefficients ranging between 0.01 to 0.56 (Stoeckli et al., 2003). Spatial and temporal measurements of swallowing kinematics can also be evaluated more objectively using specialised software and comparing measurements to norms (e.g., Kendall & Leonard, 2001), however interpretation is time-consuming particularly for the clinical setting. Methods of data extraction from radiographic images range widely (Molfenter & Steele, 2011), resulting in measurement error ranging from 2.48 to 3.06 mm (Sia, Carvajal, Carnaby-Mann, & Crary, 2012).

FEES is another commonly-used instrumental assessment, consisting of a flexible laryngoscope placed transnasally to provide a view of the hypopharynx, larynx and proximal trachea (Hiss & Postma, 2003). One of the main advantages over VFSS is that this direct visualisation allows clinicians to assess surface anatomy and secretion levels, in addition to bolus flow and structural movement. The FEES equipment is portable and can be used with patients who are otherwise unable to travel to the radiology suite, e.g., those who are ventilator-dependent or in the intensive care unit (Hafner, Neuhuber, Hirtenfelder, Schmedler, & Eckel, 2008). The procedure can be repeated often because there is no radiation exposure, and sensory testing can be carried out (Langmore, 2017). However, there are limitations to this assessment.

The oral cavity cannot be visualised and there is a brief “white-out” period during pharyngeal swallowing when the view from the laryngoscope is obscured. As with VFSS, interpretation of the dynamic images is subjective. Besides sensation, identification of the pathophysiological features underlying dysphagia can only be inferred from abnormal structural movement and bolus flow prior to the white-out period, and information after the white-out. For example, post-swallowing residue in the valleculae or pyriform sinuses after white-out could be a sign of weak pharyngeal constrictors with subsequent reduced pressure to clear residue, or uncoordinated pharyngeal contractions (Hiss & Postma, 2003). Since FEES can only visualise residue and not directly measure pressure or coordination, it cannot differentiate between possible pathophysiological features.

Pharyngeal manometry measures pressure in the oropharynx, hypopharynx and UES during swallowing, using a manometric catheter placed transnasally. Although it cannot visualise the swallowing process, manometry provides objective and quantitative information regarding amplitude and timing of pressure events. Its high temporal resolution means that it is able to detect mis-sequencing of pharyngeal pressure generation (Huckabee et al., 2014). However, shifting of anatomical structures over the low-resolution catheter because of pharyngeal shortening during swallowing can impact measurement accuracy (Huckabee, Macrae, & Lamvik, 2015). The use of manofluoroscopy, or concurrent manometry and videofluoroscopy, addresses the lack of visualisation and enables the clinician or researcher to study the relationship between visualised swallowing biomechanics and objective pressure information (Nativ-Zeltzer, Kahrilas, & Logemann, 2012). High-resolution manometry (HRM), which has 36 circumferential sensors housed in the catheter instead of the three unidirectional sensors used previously, has increased in popularity recently as it provides much more detailed information, usually presented in a three-dimensional pressure topography plot (Huckabee et al., 2015). However, highly variable pressure drift in the high-resolution catheter found during in-vivo and in-vitro studies has not been adequately corrected by standard procedures (Lamvik, Guiu Hernandez, Jones, & Huckabee, 2016). In addition, inter-rater reliability for analysis of HRM data has been found to be highly variable, with only two out of eight amplitude and duration measures having clinically acceptable levels of reliability (Lamvik, 2016). These issues have serious consequences for measurement integrity and may limit the widespread use of manometry in research and clinical practice.

3.4.3 Assessment of hyolaryngeal excursion

Given that adequate and precise movement of the hyolaryngeal complex is integral to safe, efficient and satisfactory swallowing, it is important that an accurate assessment of this component be completed on patients with dysphagia or suspected dysphagia. Impaired superior and anterior excursion of the hyolaryngeal complex can lead to airway invasion and pharyngeal residuals (Steele et al., 2011). External digital palpation of the hyoid bone and larynx has commonly been used as part of the clinical swallowing examination to assess hyolaryngeal movement during swallowing (Logemann, 1998). However, this method is highly subjective, with little evidence that it can reliably assess hyoid movement or diagnose the underlying disease pathophysiology. Intra-rater reliability for assessing hyolaryngeal elevation using digital palpation was sufficient (as measured with significant Cohen's kappa statistic) for thin liquids but not thick liquids, and inter-rater reliability was sufficient on day two of assessment but not day one (McCullough et al., 2000). Therefore, alternate methods including videofluoroscopy, sEMG, and dynamometry have been developed to assess hyolaryngeal excursion and/or its effects on swallowing biomechanics (Kuriki et al., 2012).

3.4.3.1 Videofluoroscopy

Researchers have used VFSS to measure timing (e.g., Bingjie, Tong, Xinting, Jianmin, & Guijun, 2010; Kang et al., 2010; Kendall & Leonard, 2001) and displacement of hyoid movement (e.g., Kim & McCullough, 2010; Perlman et al., 1992) in healthy controls and patients with dysphagia. There is a lack of consensus on the exact relationship between hyoid movement measured on VFSS and swallowing dysfunction. In a study with 330 patients with stroke, cancer and various neurological diseases referred for VFSS, 18.8% of all patients and 8.9% of the stroke patients were judged by two raters to have reduced hyoid elevation (Perlman et al., 1992). Reduced hyoid elevation was associated with swallowing disorders: patients with reduced hyoid elevation had 34% greater odds of oral dysphagia, and 26% greater odds of deviant epiglottic function. However, a limitation of the study was that the ratings of reduced hyoid movement were subjectively judged, and also the most difficult to agree on. Agreement between the two raters for reduced hyoid elevation was the lowest of all the VFSS measures rated, with a 70% exact agreement. The participants also had a wide range of diagnoses causing dysphagia, making it difficult to draw conclusions about diagnosis-specific swallowing disorders.

Kendall & Leonard (2001) improved on previous methods of analysing hyoid displacement by taking digitised recordings of the VFSS and making objective measurements of the distance between two frames: the hyoid at rest and at maximum displacement. They attempted to

distinguish between changes in timing and displacement of hyoid bone movement that occur as a result of pathological conditions, as opposed to those that occur in normal aging. To do this, they examined the VFSS recordings of three groups: 70 older patients (65+ years old) with nonspecific dysphagia, 60 younger (18-62 years) controls and 23 older (range 67-83 years) control participants without dysphagia. Their findings contradicted that of the Perlman et al. (1992) study, as there was no difference in hyoid displacement between older patients and older controls during swallowing of 1-mL boluses, and no difference in any hyoid timing measures between the two groups. There were significant differences between the younger and older (both control and patient) groups for most outcome measures, suggesting that differences in hyoid movement for these participants could be explained by age, rather than swallowing impairment. The significant variability in the hyoid measures for both controls and patients, as evidenced by large standard deviation values, suggests that VFSS measures of hyoid excursion and duration may not be representative of impairment or change in swallowing function over time (Molfenter & Steele, 2011). Another limitation of the Kendall & Leonard (2001) study was that patients had dysphagia due to unknown aetiology, as they were only included if they did not have a diagnosis that could cause dysphagia such as neuromuscular disease or stroke. It is possible that their dysphagia was of mild severity, which is why no differences were found between older patients and older controls, and limiting the generalisation of these results to patients with more severe dysphagia. Also, characteristics of the patients' dysphagia were not described in the study, making it impossible to define the relationship between hyoid movement and underlying pathophysiology.

Since then, research has used VFSS to investigate whether hyoid movement differs in dysphagic stroke patients with different levels of impairment, e.g., patients with and without aspiration. Results were again inconclusive, with one study noting that anterior and superior hyoid movement were not significantly different between stroke patients who aspirate and those who do not aspirate (Kim & McCullough, 2010). However, another study found that aspiration in stroke patients was associated with significantly reduced superior hyolaryngeal movement (Bingjie et al., 2010). A drawback of these studies is that aspiration status was used to divide the patients into groups. Aspiration is a symptom of swallowing impairment and can be caused by multiple underlying pathophysiological features. A better understanding of the relationship between hyoid bone movement and dysphagia would be gained by comparing groups of patients by their underlying pathophysiology, such as patients with peripheral weakness or suspected movement precision impairments, instead of aspiration status.

Paik and colleagues (2008) compared hyoid bone kinematics between healthy controls and patients that might represent different types of dysphagia. They hypothesised that patients with dysphagia due to myopathy would have weakness, while those with a central nervous system disorder (stroke) would have incoordination, with or without weakness. The two-dimensional movement of the hyoid bone was plotted frame-by-frame so that the trajectory and velocity of hyoid movement could be calculated for the duration of hyoid movement. Maximum hyoid displacements in the horizontal and vertical dimensions were not significantly different between healthy controls and stroke patients, but were reduced in myopathy patients, presumably because of weakness. However, the stroke patients demonstrated an uncoordinated pattern of movement compared to the controls, as evidenced by extraneous upwards and backwards movement of the hyoid bone and irregular movement velocities (Figure 3.2). Therefore, VFSS measurements of maximum displacement were sensitive to differentiate between healthy controls and patients with myopathy and weakness, but not stroke patients. VFSS measures of hyoid excursion are likely not sensitive enough to differentiate the numerous pathophysiological features underlying stroke, and more complex measures involving spatial and temporal aspects of movement are needed.

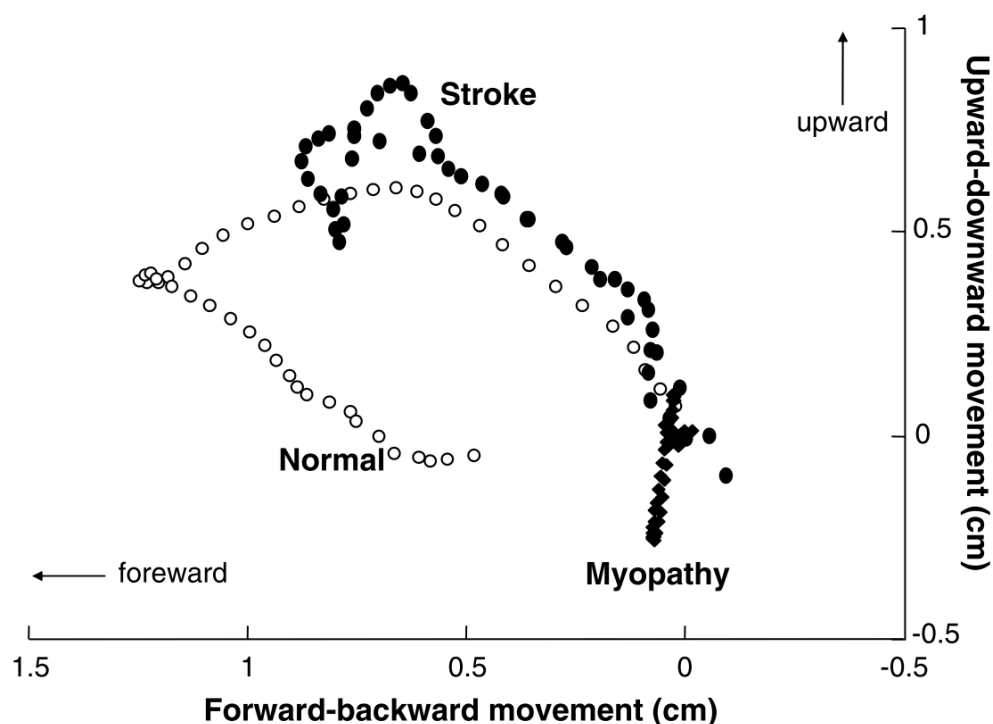


Figure 3.2. Mean kinematic trajectory of the hyoid bone (lateral view on VFSS), from onset of hyoid elevation to return to its original position. Each dot represents the horizontal and vertical displacement of the hyoid from rest position, at equal time intervals of 2% of hyoid movement

duration. A wide gap between dots indicates rapid movement, whereas dots close together indicate slow movement. From “Movement of the hyoid bone and the epiglottis during swallowing in patients with dysphagia from different aetiologies,” by N. Paik, S. Kim, H. Lee, et al., *Journal of Electromyography and Kinesiology*, Vol. 18, p. 332. Copyright 2008 by Elsevier. Reprinted with permission.

Given the drawbacks of VFSS in characterising the pathophysiological mechanisms of dysphagia, other assessment methods such as sEMG and dynamometry have been increasingly studied as evaluation practices that are more definitive of the pathophysiology underlying hyoid movement.

3.4.3.2 Surface electromyography

sEMG has been used in dysphagia research to quantify temporal and spatial aspects of muscle contraction during swallowing manoeuvres (Huckabee, Butler, Barclay, & Jit, 2005; Wheeler-Hegland et al., 2008) and swallowing of different bolus types (Ding, Logemann, Larson, & Rademaker, 2003; Leow, Huckabee, Sharma, & Tooley, 2007; Perlman, Palmer, McCulloch, & Vandaele, 1999), and to characterise neurophysiological abnormalities in patients (Ertekin, Seçil, Yüceyar, & Aydoğdu, 2004; Ertekin, Yüceyar, Aydoğdu, & Karasoy, 2001). The submental sEMG signal is sensitive to bolus consistency, with increased sEMG amplitude and duration during swallowing of thicker consistencies (Ding et al., 2003), and bolus taste, with sour and bitter boluses having greater amplitude and duration of sEMG signal respectively (Leow et al., 2007). Saliva swallowing has a longer sEMG duration than water swallows (Perlman, Palmer, McCulloch, & Vandaele, 1999).

EMG is a useful and valuable assessment tool because it is one of the few methods that can measure muscle activity during functional activities (Staudenmann, Roeleveld, Stegeman, & van Dieën, 2010). EMG signals can be detected using two methods: intramuscular and surface EMG. Intramuscular EMG uses needle or wire techniques that insert directly into the muscle. This allows for increased temporal and spatial specificity, as it measures the electrical signal from a sample of motor units directly from the source. However, intramuscular EMG is invasive, and requires specialist training to reliably place the needle or wire electrode in the desired muscle. Since it detects electrical signals from only a few selected motor units, conclusions cannot be drawn regarding overall activity of a muscle or muscle group (German, Crompton, & Thexton, 2008). On the other hand, the surface technique detects electrical activity using a surface electrode adhered to the skin over the muscles of interest. The collective

activity of several superficial muscles, such as the submental muscle group, can be detected in real-time and measured objectively. Since the submental muscles work together for hyolaryngeal excursion, measurement of overall group muscle activity using the surface technique is preferred, particularly for biofeedback (Crary & Groher, 2000; Stepp, 2012).

sEMG is particularly suited to assess the submental muscle group during swallowing because these muscles are situated relatively superficially at the floor of mouth, allowing their activity to be detected by surface electrodes. There is a significant correlation between submental muscle activity and the EMG signal measured at the skin surface (Palmer, Luschei, Jaffe, & McCulloch, 1999). Simultaneous recordings of EMG signals from the submental surface as well as directly from individual submental muscles revealed that the primary contributors to the submental sEMG reading are mylohyoid, anterior belly of the digastric, and geniohyoid muscles (Palmer et al., 1999). However, submental sEMG measurements can be influenced by lingual activity, particularly during the performance of swallowing manoeuvres such as effortful swallowing (Huckabee & Steele, 2006). Since contraction of submental muscles contributes to hyoid excursion during swallowing, research has also found strong relationships between hyoid kinematic movement and submental sEMG readings. In a study by Crary, Carnaby-Mann, & Groher (2006), young, healthy adults underwent simultaneous VFSS and submental sEMG assessment during swallowing of 5mL thin liquid. Swallowing onset, peak, and offset time points were identified for the sEMG signal and biomechanical events of hyoid movement, pharyngeal constriction, and UES opening/closing. There were strong, significant correlations between timing measures of sEMG and all three biomechanical events, although submental sEMG signals were most related to hyoid movement, illustrating the close relationship between submental muscle activity and hyoid excursion (Figure 3.3). Timing measures of maximum hyoid excursion on VFSS and maximum sEMG signal were found to be strongly correlated during both normal and effortful swallowing, but only weak-moderate correlations were reported during the Mendelsohn manoeuvre (Azola et al., 2015; Wheeler-Hegland et al., 2008).

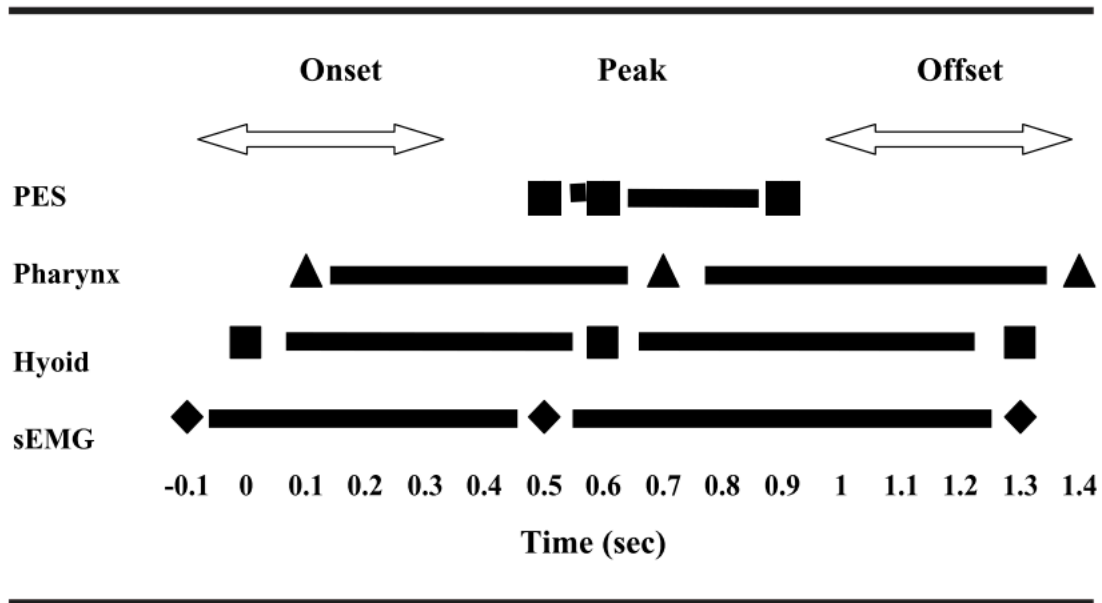


Figure 3.3. Graphical depiction of the onset, peak, and offset of the submental sEMG signal and three biomechanical swallowing events on VFSS (hyoid elevation, pharyngeal constriction, and pharyngoesophageal segment (PES) opening/closing. From “Biomechanical correlates of surface electromyography signals obtained during swallowing by healthy adults,” by M. Crary, G. Carnaby-Mann, & M. Groher, *Journal of Speech, Language, and Hearing Research*, Vol. 49, p. 190. Copyright 2006 American Speech-Language-Hearing Association. Reprinted with permission.

In addition to the temporal relationship, there is a significant spatial association between the magnitude of hyoid displacement and sEMG amplitude. Moderate positive correlations were found between maximum hyoid displacement measured on VFSS and maximum sEMG amplitude, but only after controlling for swallowing task (Wheeler-Hegland et al., 2008). Therefore, even though an association between submental sEMG with hyoid movement or submental muscle function has been reported, this relationship can differ according to swallowing manoeuvre (e.g., normal swallowing vs. Mendelsohn manoeuvre; Wheeler-Hegland et al., 2008), and possibly with age and disease (Azola et al., 2015). Studies investigating the validity of sEMG to estimate swallowing physiology have largely been completed on healthy young volunteers, limiting our understanding of the submental sEMG signal and its physiologic correlates in people with dysphagia and across the life span.

An EMG assessment protocol has been proposed as a method to characterise swallowing impairments in patients with dysphagia and differentiate between the mechanisms of dysphagia

(Ertekin, 2002). This swallowing assessment measures submental muscle activity using sEMG, laryngeal movement using a mechanical sensor placed on the midline neck between the cricoid and thyroid cartilages, and cricopharyngeus (CP) muscle relaxation using intramuscular EMG. Patients with dysphagia because of corticobulbar impairment (e.g., stroke; Ertekin, Aydoğdu, Tarlaci, Turman, & Kiylioglu, 2000) demonstrated significantly prolonged duration of submental sEMG activity, prolonged interval between onset of submental sEMG and onset of laryngeal elevation, and shortened duration of CP relaxation, when compared to healthy controls. Patients with dysphagia due to muscular disorders (e.g., myotonic dystrophy; Ertekin et al., 2001) also had significantly prolonged submental and laryngeal temporal measures, but the majority of them demonstrated normal CP relaxation timing and duration. Thus, the two groups were differentiated by assessment of CP relaxation as assessed by intramuscular EMG. However, there are limitations to this assessment protocol. The use of intramuscular EMG makes it difficult to translate this assessment into the clinical environment. Patients of different diagnoses and underlying pathophysiological features were not directly compared in the same study, and so no conclusions can be made regarding how the groups behaved relative to each other. The assessment only measured temporal coordination of swallowing events, and not spatial aspects such as amplitude of submental activity.

Submental sEMG amplitude has been used as a proxy measure of muscle force and strength, for example as an outcome measure after rehabilitation (Watts, 2013). Increased force is caused by a greater number and frequency of motor unit recruitment during muscle contraction, which in turn is reflected in increased electrical activity detected by the surface electrodes (Merletti & Parker, 2004; Stepp, 2012). However, sEMG cannot be used to directly measure muscle strength, as the exact nature of the relationship between sEMG amplitude and strength is unclear. Some research has shown that muscle activity increases linearly with greater muscle force (Stephens & Taylor, 1972); however, most studies demonstrate a non-linear relationship (Bilodeau, Schindler-Ivens, Williams, Chandran, & Sharma, 2003; Lawrence & De Luca, 1983). For example in the head and neck anatomy, while force and sEMG amplitude increase linearly for the sternocleidomastoid and semispinalis muscles, other muscle types (splenius) demonstrate a nonlinear relationship (Sommerich, Joines, Hermans, & Moon, 2000). There are many factors that affect the relationship between sEMG amplitude and muscle force, including skin fold thickness, fatigue, cross-talk, electrode placement, muscle temperature, and muscle type (Kuriki et al., 2012; Merletti & Parker, 2004). One method to reduce between-subject variability is to normalise the raw sEMG amplitude to a referent, usually the maximum

voluntary contraction during an isometric or functional task (Merletti & Parker, 2004; Stepp, 2012). However, this does not address all the factors affecting sEMG signal, such as fatigue.

In conclusion, submental sEMG is a non-invasive and relatively inexpensive technique of quantifying spatial and temporal aspects of muscle activity during swallowing and is accessible in many clinical settings. Patients can be re-assessed multiple times with sEMG without danger of radiation exposure. However, it is a non-specific measure of collective muscle contraction, cannot be used to visualise swallowing events, and does not directly measure maximum isometric strength. Further, there is high between-subject variability in raw sEMG amplitude, necessitating normalisation to a referent. Therefore, submental sEMG is best used as a clinical tool to measure the relative spatiotemporal characteristics of muscle activity during functional swallowing tasks, particularly in conjunction with other dysphagia assessments.

3.4.3.3 Dynamometry

Dynamometry has been commonly used to obtain objective and reliable measures of muscle force during strength assessment of the upper and lower extremities (Bohannon, 1986). It is much more difficult to access the muscles in order to measure the force used for swallowing because they are smaller than limb muscles and overlap each other. Researchers have developed a jaw-opening force test to measure the maximum isometric force (in kg or newtons) of the submental muscles during jaw opening, using a small dynamometer placed under the chin and secured to the head (Figure 3.4; Tohara et al., 2011). Participants were asked to open the jaw widely and with as much strength as possible against the dynamometer. Since many of the submental muscles that elevate the hyoid are also involved in opening the jaw (mylohyoid, anterior belly of the digastric, and geniohyoid), measurement of submental muscle force during jaw opening was proposed to provide insight into submental muscle strength and hyolaryngeal excursion during swallowing. Measurement of jaw-opening force with dynamometry has been used to quantify changes in submental muscle strength in healthy individuals (Iida et al., 2013; Machida et al., 2017; Shinozaki et al., 2017), screen for dysphagia (Hara et al., 2014), and as an outcome measure after rehabilitation (Kraaijenga et al., 2015; Matsubara et al., 2018; Wada et al., 2012). Advantages of dynamometry are that it can be repeated many times, it is non-invasive and portable, and it provides objective and measurable data regarding muscle strength.

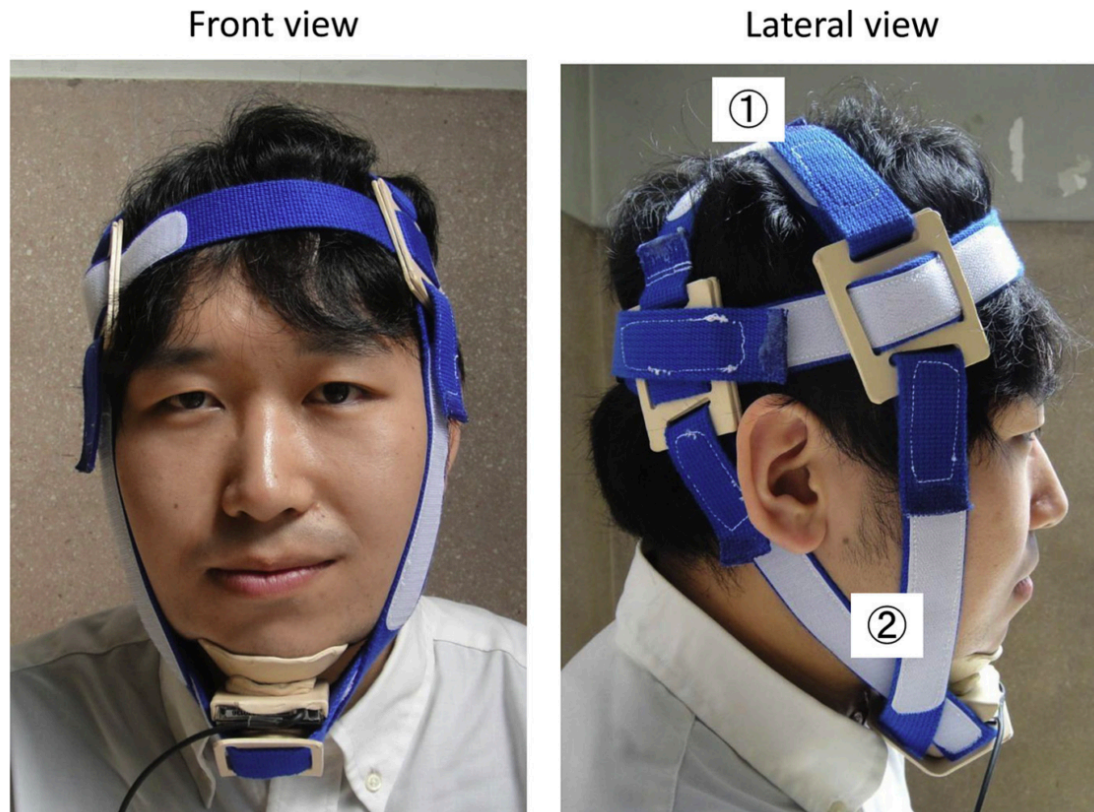


Figure 3.4. Jaw-opening sthenometer. The jaw is kept in a fixed position using adjustable straps that cross on the top of the head (1), with the dynamometer secured under the chin (2). From “Jaw-opening force test to screen for dysphagia: Preliminary results,” by K. Hara, H. Tohara, et al., *Archives of Physical Medicine and Rehabilitation*, Vol. 95, p. 869. Copyright 2014 by Elsevier. Reprinted with permission.

Studies have examined the relationship between jaw-opening force and swallowing function, but understanding of this relationship is still limited. Iida and colleagues (2013) found that both age and sex were significant factors in jaw-opening strength. Younger participants (aged below 70 years) had significantly greater jaw-opening force than older participants (over 70 years of age), while men had significantly greater jaw-opening force than women. Mean jaw opening force of healthy elderly adults (7.8 ± 3.0 kg; Iida et al., 2013) is higher than that of elderly patients with dysphagia (4.95 ± 2.93 kg; Hara et al., 2014). Submental muscle size appears to be related to jaw-opening force (Kajisa et al., 2018). The cross-sectional area of the geniohyoid muscle, as measured using ultrasonography, was significantly and positively associated with jaw force, although there was no relationship between the cross-sectional area of the anterior belly of the digastric and jaw force. The mylohyoid was too thin to be measured using

ultrasonography. Skeletal muscle mass of the body (normalised to body height) was also significantly and positively associated with jaw force, but in women only (Kajisa et al., 2018).

Researchers also hypothesised that jaw-opening force would be related to hyoid position and displacement on VFSS, because of the role the submental muscles play in superior and anterior excursion of the hyoid (Shinozaki et al., 2017). Contrary to expectations, there were no significant correlations between jaw force and any measure of hyoid position or displacement in healthy women. In the healthy male participants, there was a strong and positive correlation between jaw-opening force and resting hyoid position (measured as the vertical distance from the hyoid to the x-axis of a coordinate system, where the y-axis was aligned with the cervical vertebrae and the x-axis was perpendicular to the y-axis), but moderate-strong negative correlation between jaw-opening force and hyoid displacement measures. In other words, male participants with lower jaw force had a lower resting hyoid position but increased hyoid displacement during swallowing. This suggests that healthy elderly individuals have an optimal maximum hyoid position during swallowing that is unaffected by jaw strength, and healthy elderly men compensate for a lower resting hyoid position by increasing the hyoid displacement during swallowing to reach this optimal maximum hyoid position. In this study, only healthy participants with intact swallowing were evaluated. It is unknown whether jaw-opening force affects hyoid kinematics differently in patients with dysphagia. Future research investigating submental muscle strength should include both swallowing and non-swallowing tasks, to gain a better understanding of submental muscle function during both maximal and submaximal behaviours.

Researchers from the same group investigated the relationship between jaw-opening force and swallowing impairment in dysphagic patients, and assessed whether the jaw-opening force test could predict aspiration and pharyngeal residue that were observed during FEES (Hara et al., 2014). The jaw-opening force test had high sensitivity and specificity (over 0.8) for predicting residue in the valleculae and pyriform sinuses, but low sensitivity (0.57 and 0.93 for men and women respectively) and specificity (0.79 and 0.52) to aspiration. Measurement of jaw-opening force as an assessment of suprahyoid muscle strength may be a useful tool to objectively identify those with and without impaired swallowing strength, but it was not sufficiently sensitive to identify all patients with dysphagia. This suggests that patients may have other types of impairment (besides weakness of the suprahyoid muscles) causing swallowing impairment. A comprehensive assessment of the underlying mechanisms of

dysphagia might include measurements of swallowing movement precision, in addition to measurements of swallowing strength, of the submental muscles.

3.5 Treatment of dysphagia

The goals of dysphagia management are to maintain or improve nutritional and hydration status, ensure pulmonary safety, rehabilitate swallowing ability to pre-morbid level, and improve overall quality of life. Unfortunately there is insufficient evidence for dysphagia interventions, as concluded by a Cochrane review of randomised controlled trials in dysphagic stroke patients (Geeganage, Beavan, Ellender, & Bath, 2012). Typical dysphagia therapies used after stroke can be divided into two categories: compensatory and rehabilitative treatments.

3.5.1 *Compensatory strategies*

Compensatory strategies, including diet modification, postural changes, and non-oral feeding techniques, do not aim to change the physiology of swallowing in the long-term, but instead promote immediate change in bolus flow. When the compensatory strategy is being implemented, the signs and symptoms of dysphagia are reduced with immediate effect, however this effect is discontinued when the strategy is removed (Daniels & Huckabee, 2014; Welch, Logemann, Rademaker, & Kahrilas, 1993). Diet modification (such as thickening liquids and pureeing solids) and postural changes (such as chin tuck or head turn) changes the speed and direction of bolus flow, thus reducing post-swallow residuals and airway invasion (Bülow, Olsson, & Ekberg, 2003; Logemann, Kahrilas, Kobara, & Vakil, 1989; Shanahan, Logemann, Rademaker, Pauloski, & Kahrilas, 1993). Non-oral feeding using nasogastric (NG) or percutaneous endoscopic gastrostomy (PEG) tubes allow circumvention of the disordered swallowing mechanism. Nutrition and hydration are delivered via an alternate route, with the goal of reducing aspiration and aspiration pneumonia, especially in the acute phase (Dennis, Lewis, Cranswick, & Forbes, 2006). However, there are many significant disadvantages to compensatory strategies that should be considered prior to use. There is conflicting evidence as to whether non-oral feeding can reduce pulmonary complications and mortality, and feeding tubes have been found to cause medical complications including infection, tube dysfunction, and even aspiration pneumonia (Anis et al., 2006; Langdon, Lee, & Binns, 2009). Patients may have reduced acceptance of modified foods and drinks at mealtimes, resulting in reduced caloric intake and dehydration (Cichero, 2013). Postural changes require the patient to have adequate cognition, attention and adherence to perform the postures consistently, since the effect disappears after the posture is removed (Rasley et al., 1993). There is also evidence that

the effects of various postures is different between individuals (Chaudhuri, Brady, & Ng, 2013; Shanahan et al., 1993), so these strategies should not be applied without instrumental assessment.

3.5.2 Strength-based rehabilitation

Compensatory strategies can be beneficial for short-term management (Bülow et al., 2003; Daniels & Huckabee, 2014). Rehabilitation exercises on the other hand, if based on an understanding of neuroplasticity and the underlying pathophysiology of dysphagia, can promote long-term change. Most rehabilitation techniques are strength-based, because of the assumption that dysphagia is caused by weakness in the swallowing mechanism. Two of the most common rehabilitation exercises, effortful swallowing and the Mendelsohn manoeuvre, evolved from being strength-based compensatory strategies. Other strength-based swallowing exercises include head lifts (Shaker et al., 2002), expiratory muscle strength training (EMST; Wheeler, Chiara, & Sapienza, 2007; Wheeler-Hegland, Rosenbek, & Sapienza, 2008), tongue-holding or Masako manoeuvre (Fujiu & Logemann, 1996), and lingual resistance exercises (Robbins et al., 2007).

The effortful swallowing technique is commonly used to improve muscle strength and swallowing function. Research has found that individuals are able to modulate their motor response to increase effort during swallowing. This technique is often taught to patients by clinicians as a compensatory strategy to increase pressure on the bolus as it passes through the aerodigestive tract, thereby reducing pharyngeal residual after swallowing and preventing airway invasion (Lazarus, Logemann, Song, Rademaker, & Kahrilas, 2002; Logemann, 1998). Compared to non-effortful or “regular” swallowing, effortful swallowing has been found to increase the duration of the pharyngeal response, maximum hyoid anterior excursion, laryngeal closure, UES opening, and total swallowing duration, as seen on VFSS, and increase oral pressures (Hind, Nicosia, Roecker, Carnes, & Robbins, 2001). In addition, investigation of the effects of effortful swallowing on manometric pharyngeal pressures and submental sEMG muscle activity have demonstrated greater submental muscle contraction, increased pharyngeal pressures, and decreased pressure at the UES during effortful swallowing (Hiss & Huckabee, 2005). Previous research has demonstrated that sEMG activity during normal swallowing is approximately 42 – 47% of the maximum muscle activity generated during effortful swallowing (Huckabee et al., 2005; Wheeler-Hegland et al., 2008; Yeates, Steele, & Pelletier, 2010).

Although most studies have focused on effortful swallowing as a compensatory strategy for impaired swallowing, there has been some research on effortful swallowing as a rehabilitation exercise (Bryant, 1991; Huckabee & Cannito, 1999; Li et al., 2016; Lin et al., 2003; Park et al., 2009; Zhang, Huang, Wu, Chen, & Huang, 2014; Zhen, Wang, Tao, Wang, & Chen, 2012). Studies have demonstrated improvements in bolus flow on VFSS, oral diet tolerance, and pulmonary status in dysphagic patients, as a result of intensive rehabilitation programs that included effortful swallowing paired with biofeedback (Bryant, 1991; Huckabee & Cannito, 1999). The majority of patients were able to have their feeding tubes removed and return to a full oral diet. However, since these rehabilitation programs used effortful swallowing with biofeedback, and incorporated additional exercises other than effortful swallowing in the protocol, it is unknown whether improvement was due to the effect of effortful swallowing in isolation. Other studies have demonstrated that effortful swallowing exercise on its own does not result in significant physiological or functional change, but needs to be performed with biofeedback (Li et al., 2016) or against resistance provided by electrical stimulation (Park et al., 2009) to improve swallowing. Stroke patients with dysphagia who participated in effortful swallowing training with game-based biofeedback had significantly increased hyoid displacement and fewer oral diet restrictions after training, while patients who completed effortful swallowing without biofeedback had no change (Li et al., 2016). Although a biofeedback target was provided to the patient to encourage increased effort and muscular recruitment during exercise, the target threshold and the rules for when to progressively increase the target were not specified (Bryant, 1991; Huckabee & Cannito, 1999; Li et al., 2016). This makes it difficult for researchers and clinicians to replicate the exercise procedure.

Little is known about the appropriate dosage and intensity for effective strength training in swallowing; however, principles of effective exercise can be inferred from the fields of physical rehabilitation and sports training. In order to improve the force-generating capacity of a muscle and for central and peripheral adaptation to occur, the exercise task must be completed at an intensity that exceeds the muscle's usual level of activity. The physiologic load placed on the muscle should be higher than the usual demand and progressively increase over time (Burkhead et al., 2007; Patten et al., 2004). Generally, the target load is calculated as a percentage of the maximum force generated in a single repetition (also called a 1-repetition maximum or 1-RM). Exercise physiologists recommend an initial target goal of at least 60% of 1-RM but preferably higher (Porter, 2000). The most beneficial range has not been elucidated for dysphagia rehabilitation. Previous research on progressive strength-training exercises in swallowing have utilized target training loads at 60-80% of 1-RM (Kim &

Sapienza, 2005; Robbins et al., 2007). These progressive resistance training programs have demonstrated improved physiological and functional outcomes in patients with dysphagia (Robbins et al., 2007; Troche et al., 2010).

However, researchers have raised some concerns regarding the efficacy and appropriateness of strength training. While strength training has been shown in the limb literature to be effective at increasing muscular strength, the evidence that increased strength generalises to improved participation and performance in functional daily activities is inconclusive (Harris & Eng, 2010; Latham, Bennett, Stretton, & Anderson, 2004; Patten et al., 2004; Rasch & Morehouse, 1957; Symons, Vandervoort, Rice, Overend, & Marsh, 2005; Van Peppen et al., 2004). Furthermore, strength training may have adverse effects, including fatigue (Ament & Verkerke, 2009), increased muscle tone (Clark, 2003), and detraining effects when strength training is stopped (Baker, Davenport, & Sapienza, 2005; Clark, O'Brien, Calleja, & Corrie, 2009). In the dysphagia literature, some studies have demonstrated that effortful swallowing does not improve airway invasion or amount of pharyngeal residual in patients with dysphagia (Bülow, Olsson, & Ekberg, 2001), but can cause reduced anterior hyoid movement and laryngeal elevation (Bülow, Olsson, & Ekberg, 1999), raising concern that this may negatively affect airway closure. Another adverse effect was noted in a case study by Garcia and colleagues (2004), who found that effortful swallowing resulted in premature tongue base-to-posterior pharyngeal wall contact and re-direction of the bolus through the nasal cavity.

Huckabee & Steele (2006) investigated these discrepancies and found that the instructions given to participants on how to produce an effortful swallow can affect the resulting pharyngeal pressure dynamics. Healthy participants who used a technique that emphasised tongue-to-palate pressure (as opposed to completing an effortful swallow *without* emphasising tongue pressure) generated increased oropharyngeal pressure and submental sEMG activity. These studies reinforce the notion of prescribing rehabilitation strategies that are specific to the patient's underlying disorder and pathology. Patients with vallecular residuals as a result of poor tongue base retraction might benefit from the increased pressure generated by an effortful swallow completed with tongue-to-palate pressure. However, this treatment may be contraindicated for those demonstrating vallecular residual because of reduced anterior hyoid movement, as effortful swallowing may exacerbate this underlying impairment (Bülow et al., 1999).

The inconsistencies in the corticospinal and corticobulbar literature regarding the benefits of strength training may be explained by heterogeneity in the patients' severity and type of impairments, particularly after stroke (Patten et al., 2004). Not all patients after stroke experience weakness, and the level of severity is different between patients (Bohannon, 2007). In a study comparing different types of upper extremity training after stroke, patients were stratified by stroke severity. Only the patients in the "less severe" group demonstrated any effect of the exercise program, while the "more severe" patients had no improvement (Winstein et al., 2004). It is important to identify the pathophysiology of impairment before prescribing treatment, as the effectiveness of the training program will likely be different depending on the type and severity of impairment. For a patient whose swallowing impairment is not characterised by weakness, training which aims to increase muscular strength would likely be ineffective. Given that swallowing requires precise timing and accuracy of movement, instead of maximum force, researchers have begun investigating skill-based training as an alternative approach to rehabilitation.

3.5.3 Shift to skill-based training

The effects of skill training have been researched extensively in the limb literature, but only recently have preliminary studies on skill-based training in dysphagia emerged. Most of our understanding of the principles of skill-based training is based on these limb studies. Motor skill learning is defined as "increasing spatial and temporal accuracy of movements with practice" in order to reach a movement goal (Willingham, 1998). Skill acquisition involves improved precision of performance by decreasing errors, while maintaining movement speed (Kitago & Krakauer, 2013). While strength training improves the ability to generate force through resistance exercise, skill training focuses on the acquisition and refinement of movement sequences to improve the accuracy of motor execution (Adkins, Boychuk, Remple, & Kleim, 2006; Kitago & Krakauer, 2013).

Extensive practice is a key component of motor skill learning, but simply practising the same movement repeatedly is not enough to affect long-lasting change at the behavioural or neural level (Plautz, Milliken, & Nudo, 2000). Instead, varying the level of task difficulty during practice can improve the retention and generalisation of learned skills to new tasks (Krakauer, 2006). Giving individuals tasks that are slightly more difficult than their current level of performance, and continually increasing this difficulty when they have mastered the current level, improves both learning and motivation (Green & Bavelier, 2008). Another principle of skill training is the use of extrinsic or augmented feedback, which provides external

information to the participant regarding their patterns of movement (knowledge of performance) or whether they achieved their target (knowledge of results; Kitago & Krakauer, 2013). Augmented feedback can help patients to cognitively problem solve their errors and develop strategies to improve skill acquisition. Finally, skill learning is generally most efficient when the training protocol is similar to the target behaviour, illustrating the specificity of learning principle (Green & Bavelier, 2008).

The changes seen in cortical areas after skill training reinforces the strong association between skilled movement and cortical control. Compared to unskilled or passive strength training, skill training in the limbs has been found to result in increased corticospinal excitability (Jensen, Marstrand, & Nielsen, 2005) and plasticity of the motor cortex (Kleim, Barbay, & Nudo, 1998) in healthy subjects. Skill training is also associated with neuroplastic change in the lesioned cortex of stroke patients. Stroke patients who were randomly assigned to skilled, task-specific training with their hemiparetic arm demonstrated a more normal pattern of brain activation in the contralesional hemisphere, compared to those who received general non-specific training (Boyd, Vidoni, & Wessel, 2010). In another study, stroke patients had improved motor functioning, as evidenced by decreased time to complete a fine motor task, and greater motor cortex representation of the hand measured with transcranial magnetic stimulation, immediately after dexterity training of the hemiparetic hand (Liepert, Graef, Uhde, Leidner, & Weiller, 2000).

There has been limited research on the translation of skill training and motor learning principles to swallowing rehabilitation. It was found that a tongue-pressure training protocol using both strength targets (maximum isometric pressure) as well as accuracy targets (variable targets at 20-90% of maximum isometric pressure) resulted in increased tongue pressure, improved tongue pressure generation accuracy, reduced airway invasion, and improved bolus control on videofluoroscopy (Steele et al., 2013; Yeates, Molfenter, & Steele, 2008). However, since patients were trained on both strength and skill targets, the effects of skill training in isolation on this corticobulbar task are unknown, and likely were confounded by strength increases.

The effectiveness of skill training using a novel treatment protocol, Biofeedback in Strength and Skill Training (BiSSkiT), was investigated in patients with Parkinson's Disease (Athukorala et al., 2014). Submental sEMG was used to measure the timing and amplitude of muscle activity, which was displayed on a computer screen as biofeedback (Figure 3.5). A square target was placed on the screen, with instructions for the participant to swallow so that

the peak of the waveform fell within the square. The amplitude range of the computer screen was calibrated to the patient's maximum muscle activity during five effortful swallows performed at the beginning of each session, so the task was submaximal and could be completed within the patient's available strength level. The target moved to a random vertical and horizontal location at each trial, getting progressively smaller in size as the patient became more proficient at the task, requiring increased spatiotemporal precision of swallowing.

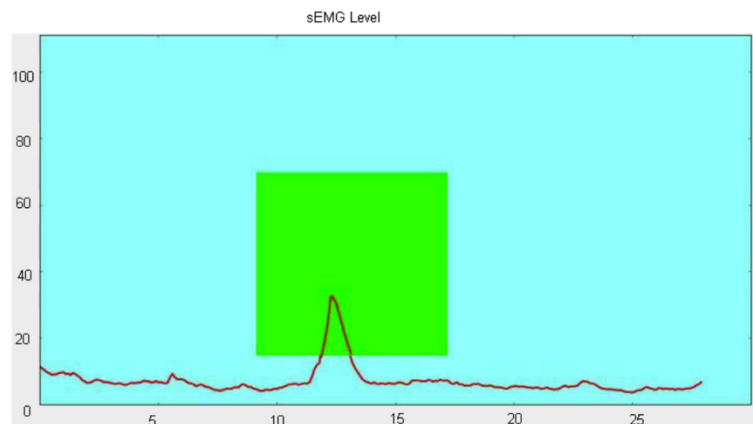


Figure 3.5. Visual biofeedback of submental sEMG activity (waveform) and skill-training target (green square) in BiSSkiT software. From “Skill training for swallowing rehabilitation in patients with Parkinson's disease,” by R. Athukorala, R. Jones, et al., *Archives of Physical Medicine and Rehabilitation*, Vol. 95, p. 4. Copyright 2014 by Elsevier. Reprinted with permission.

Results demonstrated that immediately after skill training, the patients demonstrated decreased time per swallow during the timed water swallow test and reduced sEMG durational measures of premotor and preswallow time, suggesting more efficient timing and speed of movement (Athukorala et al., 2014). Carryover effects were seen from dry swallows to untrained water swallows. Outcome measures did not change in the two weeks after conclusion of treatment, indicating retention of skill training effects. Swallowing-related quality of life also demonstrated improvement. The authors concluded that the varying targets and progressively challenging levels contained in the training protocol provided the opportunity for patients to increase conscious control of their swallowing force and timing. For a behaviour like swallowing that produces minimally observed external movement, it can be difficult for patients to evaluate whether their movement pattern matches the intended motor plan and decide which corrections are needed. Patients in this study were able to use biofeedback to

integrate information about their errors and problem-solve towards a more accurate sEMG response on the next trial. The BiSSkiT software used precision and accuracy targets at 20-70% of maximum muscle activity (Sella, 2012). Setting the upper range of the targets at a submaximal 70% meant that the task would not be confounded by any effect of strengthening during effortful swallowing, and only movement precision training would be targeted. However, it is unknown whether setting the low boundary at 20% of maximum contraction is appropriate, since no study has investigated the functional range of muscle activity used for swallowing, particularly the minimum amount of muscle activity needed.

The advent of skill training for swallowing is an acknowledgement that dysphagia might be caused by pathophysiological features other than weakness (Huckabee & Macrae, 2014). While skill-based training has demonstrated potential in the rehabilitation of patients with swallowing impairments, little is known about which patients can benefit the most from skill training. One limitation of previous research in both the corticobulbar and corticospinal literature is that the prescription of strength and skill training continues to lack specificity. For example, patients are randomly assigned to strength or skill training without first assessing or controlling for the underlying pathophysiology (such as weakness, reduced dexterity, or spasticity) of their motor impairment (Athukorala et al., 2014; Boyd et al., 2010; Liepert et al., 2000; Steele et al., 2013). It is difficult to assess the efficacy of a rehabilitation technique if the sample has heterogeneous baseline impairments. Not all stroke patients have strength deficits (Bohannon, 2007), and similarly, not all stroke patients may have impairments in movement precision. It is likely that strength training for the limbs is more effective for those with strength impairments, and skill training for those with movement accuracy or precision impairments. This can be applied to swallowing behaviours: if strength training is centred on the assumption that dysphagia is caused by weakness at the peripheral level, then skill training assumes that dysphagia is caused by decreased execution of coordinated, goal-directed movement at the central level (Huckabee & Macrae, 2014). Further research is required to separate the possible contributing factors to the underlying pathophysiology of dysphagia, and to investigate the differences between strength and movement precision impairments in swallowing.

Chapter 4. Clinical measurement of strength and skill

4.1 Definitions

Strength is defined as the capacity of a muscle or muscle group to generate force for initiation and maintenance of movement, while muscular weakness is the deficiency in generating adequate force (Ng & Shepherd, 2000). The definition of weakness used in this document comprises decreased force generation that may be caused by multiple mechanisms, including direct changes to agonist motor units, secondary adaptive changes to muscle fibres, and indirect antagonist restriction of agonist activation (Bourbonnais & Vanden Noyen, 1989; Ng & Shepherd, 2000). In the limb literature, dexterity is the ability to precisely and quickly coordinate voluntary movement in a spatiotemporal task (Canning et al., 2000). A related concept is that of motor skill, which has been defined as an acquired, fixed sequence of voluntary movements that are accurately coordinated in time to achieve a goal (Hikosaka, Nakamura, Sakai, & Nakahara, 2002). Given that swallowing is a precise, coordinated sequence of movements, the concepts of dexterity and motor skill may relate to swallowing behaviour. However, since swallowing behaviour is not entirely voluntary, it is proposed that the term movement precision be used instead to refer to coordination in swallowing. In this document, movement precision in swallowing is defined as the spatial and temporal accuracy of movement to meet environmental demands.

While this document will focus on classifying patients based on movement precision and strength deficits, it is important to distinguish between absolute performance level (motor execution) and the ability to acquire and retain new skills (motor skill learning; Raghavan, 2007). While execution deficits of strength and movement precision are commonly noted after neurological damage, it is unclear whether motor learning is also affected. The limited research in this area suggests that stroke patients have intact motor learning despite impaired motor execution (Kitago & Krakauer, 2013). Stroke patients demonstrated significantly impaired skill execution, as evidenced by large errors during a visuomotor task, as well as poor isometric strength, but their rate of learning over eight training trials did not differ from healthy controls (Van Hedel et al., 2010). This thesis will focus on exploring the concepts of strength and movement precision impairment as deficits of motor execution.

4.2 Impaired strength and skill in corticospinal muscles

In the limb literature, deficits in strength and skill are considered to be the major contributors to impaired functioning after brain damage, with the two factors accounting for 71% of the variability in function for the first six months post stroke onset (Canning et al., 2004). Given the importance of strength and skill performance to functional behaviours in the limb, research has focused on methods of quantifying and discriminating between strength and skill

impairment in patients after neurological damage. It is challenging to determine whether functional impairments are a result of decreased strength, decreased skill, or both, because most complex tasks require aspects of skill and strength. For example, reaching tasks need both adequate muscle force to move the arm against gravity, as well as adequate skill to coordinate the timing and precision of angular motions of the shoulder, wrist, and fingers.

Studies investigating the independent contributions of strength and skill have addressed this issue by measuring strength as isometric force during maximum voluntary contraction, and measuring skill during visuomotor tracking tasks which can be completed with minimal strength (Ada, O'Dwyer, Green, Yeo, & Neilson, 1996; Canning et al., 2004, 2000). One visuomotor tracking task required participants to track a randomly moving target on a display by controlling the speed, timing, and amplitude of their elbow flexor and extensor movement (Figure 4.1). Motor skill was quantified by calculating the coherence square function using cross-correlational and spectral analysis, which measured the proportion of response that was correlated with the target during the visuomotor task. This assessment simulated the complex skilled movement needed in activities of daily living, by incorporating sensorimotor integration, motor planning and execution, and error correction required in skilled tasks. The strength requirement was minimal since the elbow was supported against gravity, the required elbow movement was within the optimal mid-range, and very little muscle activation was needed to produce a response. Therefore, performance on the task was thought to be attributed only to the patients' available skill level. However, a certain amount of strength was still needed, and it is possible that patients with lower levels of strength needed more effort to complete the skill task compared to those with intact strength (Milot, Nadeau, Gravel, & Requiaio, 2006).

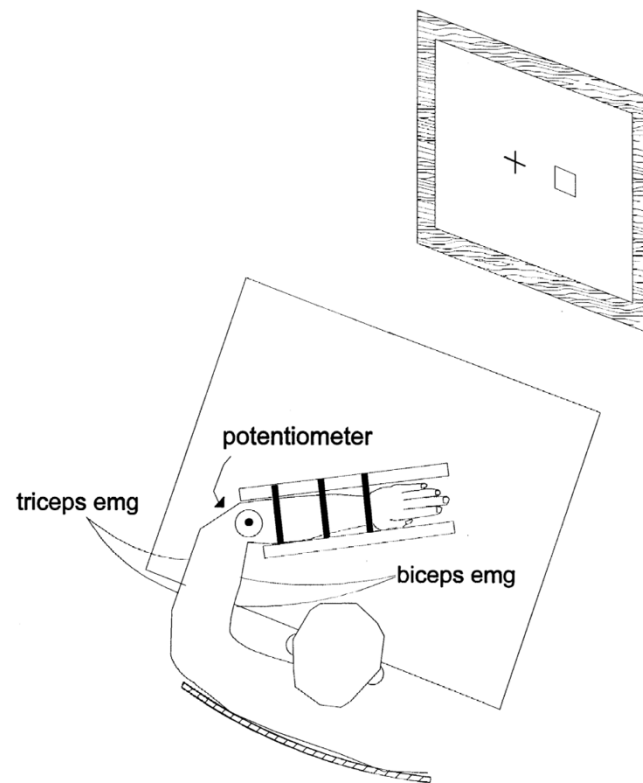


Figure 4.1. Visuomotor tracking task for measuring elbow dexterity. The forearm and elbow are supported so that rotation at the elbow joint results only in elbow extension (moves cursor to the left) and flexion (moves cursor to the right). Patient controls response cursor on the screen (cross) so that it moves into the target (square). From “Abnormal muscle activation characteristics associated with loss of dexterity after stroke,” by C. Canning, L. Ada, and N. O’Dwyer, *Journal of the Neurological Sciences*, Volume 176, p. 48. Copyright 2000 by Elsevier. Reprinted with permission.

Another method of assessing motor skill after stroke controlled for strength by calibrating the visuomotor task to each participant’s strength level, which was quantified as the maximum voluntary contraction (MVC) of ankle flexion (Van Hedel et al., 2010). A red biofeedback line moving from the left to the right of the screen was controlled by the participant, moving up with dorsal flexion and down with plantar flexion. Participants controlled timing and amplitude of their dorsal and plantar flexion to match the target trajectory line (in blue) as accurately as possible. The target trajectories contained submaximal target levels of dorsal and plantar flexion torques at 20, 40, 60 and 80% of each participant’s MVC, so that any weakness would not interfere with performance of the skilled task. Skill was measured as the error (root mean square) between the response and target trajectories; increased skill was represented by a small error and vice versa.

Using the above methods, studies have compared strength and skill impairments of the limbs in patients with lesions of the corticospinal tract at the spinal level (spinal cord injury; SCI) versus cortical level (stroke; Tomita & Usuda, 2013; Van Hedel et al., 2010). Researchers hypothesised that stroke and SCI patients would have comparable strength impairments because weakness can be caused by both UMN and LMN damage. However, it was expected that the cortical damage seen in stroke would result in skill impairments in stroke patients only. Since the cortical areas in SCI patients are spared, they would have relatively intact skill within the confines of their peripheral weakness. Results demonstrated that strength was similarly impaired in both the SCI and stroke patients. However, while ankle dexterity in the SCI patients was intact (no significant difference in visuomotor tracking accuracy or temporal coordination between SCI and healthy control group), ankle skill for the stroke group was significantly diminished in not only the hemiparetic leg (Tomita & Usuda, 2013; Van Hedel et al., 2010) but also the non-paretic side (Van Hedel et al., 2010). A deterioration in skilled, coordinated movement was associated with supraspinal lesions only, and can manifest regardless of the presence of weakness. Therefore, strength and skill deficits can occur separately from each other, and depends on the anatomical location of the lesion.

Research has found that performance on tests of strength in stroke patients was poorly correlated with performance on skill tests (Ada et al., 1996). This reinforces that strength and skill can be differentially affected by stroke, and stroke patients may have varying levels of strength and skill impairment. Another study was undertaken by the same research group to further investigate the mechanism and characteristics of skill impairment after stroke (Canning et al., 2000). Sixteen stroke patients (patients with spasticity were excluded) and 10 healthy controls participated in a visuomotor tracking task using elbow flexion and extension. The patients were divided into a low and high dexterity group based on spatial and temporal performance during the visuomotor tracking task. It was found that there were no significant differences between the low and high dexterity groups in terms of maximum EMG speed and co-activation of the agonist and antagonist muscles as measured by EMG during tracking, suggesting that loss of dexterity is not characterised by abnormal speed or co-activation. Weakness was not a contributing factor because minimal elbow strength was required to control the response cursor and participate in the task. The low dexterity group did demonstrate excessive agonist muscle activation compared to the high dexterity patient group and healthy controls. This was unlikely due to spasticity because patients with spasticity were excluded, and the maximum speed required for the dexterity task was not fast enough to cause velocity-dependent stretch reflexes. Therefore, impaired dexterity in these stroke patients was not

related to slowness, abnormal co-contraction, weakness, or spasticity, but instead was associated with excessive and imprecise muscle activation, with the patient unable to skilfully modulate the amount and timing of muscle activation to meet environmental needs. This study lends support to the idea that dexterity deficits can manifest separately from other motor impairments such as strength. It is important for both strength and skill to be assessed after stroke, as different treatment options will be needed depending on the patient's impairment profile. However, little is known about strength and skill impairments in other behaviours after stroke, particularly in the corticobulbar system.

4.3 Impaired strength and skill in motor speech

Studies in the motor speech literature may shed some light on the relevance of skill and strength in the healthy and damaged corticobulbar system. It is well-established that speech is a submaximal task, since production of accurate and fast speech movements requires less than the maximum force generated by the oral musculature (Bunton, 2008; Bunton & Weismer, 1994). Healthy individuals use approximately 20 – 25% of their maximum force of the oral musculature to produce speech (Neel, Palmer, Sprouls, & Morrison, 2015). It has been assumed that oral-motor strength is an important contributor to speech production, with subjective measurements of strength (e.g., tongue push against clinician-provided resistance) being a dominant part of motor speech evaluations (Duffy, 2005). However, a review of the literature concluded that there was insufficient evidence for a relationship between jaw, lip, and tongue force and measures of speech production (speech intelligibility and severity scores) in patients with neurological disorders (Bunton, 2008). Tongue strength has been found to be a poor predictor of diadochokinetic rates and articulation rates during reading for healthy speakers (Neel & Palmer, 2012). Although patients with muscle atrophy from OPMD had less than half of the maximum tongue strength of healthy controls, they did not have substantially impaired acoustic measures of speech and voice production nor reduced ratings of speech intelligibility (Neel et al., 2015). Since muscle strength is not the only factor contributing to the complex behaviours of speech in both the healthy and patient population, researchers recommend that focus should also be placed on other aspects such as movement skill and coordination, in order to fully understand motor speech impairments (Neel & Palmer, 2012).

Research has investigated whether visuomotor tracking tasks can assess and quantify the accuracy and precision of movement in different speech systems, and whether these tasks can be used to differentiate between healthy and disordered groups (Ballard, Robin, Woodworth, & Zimba, 2001; McClean, Beukelman, & Yorkston, 1987). Ten healthy participants and six

patients with dysarthria secondary to various neurological aetiologies tracked a sinusoidal target displayed on a screen by controlling the spatiotemporal movement of various speech muscles. Lip and jaw performance were tested using strain gauges attached to the lower lip and underside of the jaw. Laryngeal performance was measured as fundamental frequency during sustained phonation, while respiratory performance was assessed by air pressure changes using a face mask. Healthy participants demonstrated good performance on the tracking task, as evidenced by consistently high correlation values between the target waveform and their response (Figure 4.2A). On the other hand, the participants with dysarthria had overall impaired tracking performance compared to the healthy controls, and demonstrated wide variability in performance (Figure 4.2B; McClean et al., 1987). Patients with Parkinson's Disease had relatively normal performance on jaw tracking, but were impaired in lip, respiratory, and laryngeal tracking. Compared to the other patients with dysarthria, the patient with lacunar infarcts had the most difficulty tracking with the larynx, but had unaffected performance with the lip and jaw. This illustrates that not only does skilled performance of the oral musculature contribute to speech production, various speech subsystems can demonstrate differential levels of skilled performance.

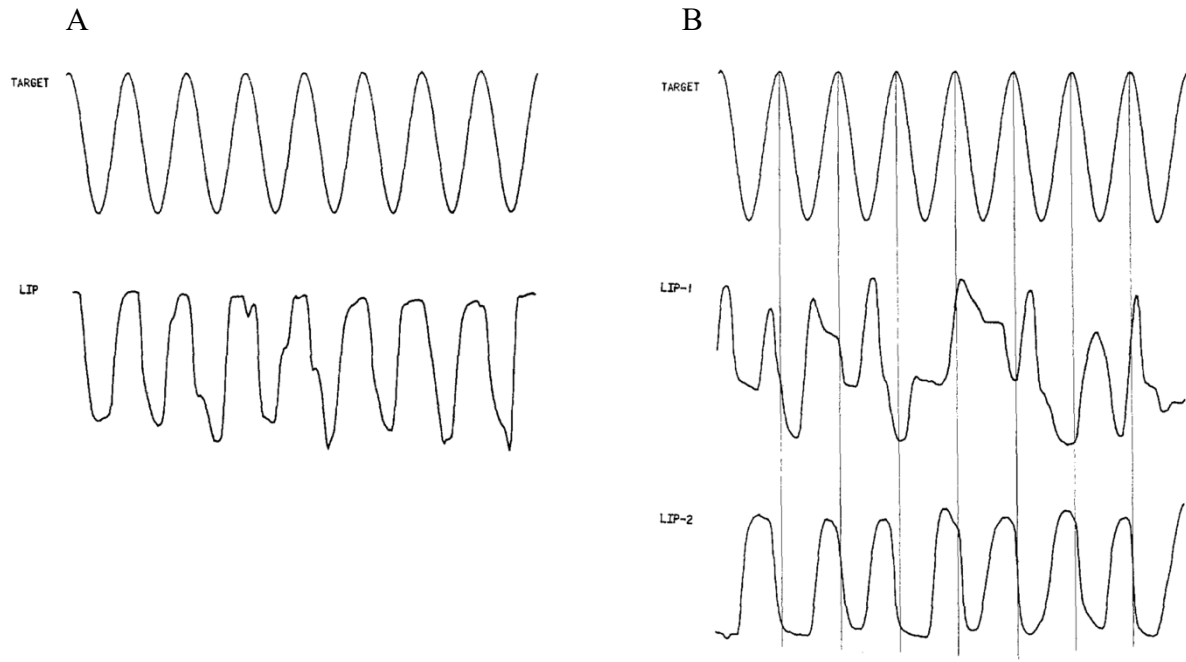


Figure 4.2. Top waveforms represent target signal, with lip tracking performance represented by waveforms below. A: Healthy participant; B: Patient with Friedrich's ataxia at two different times, indicating variability in tracking performance. From "Speech-Muscle Visuomotor

Tracking in Dysarthric and Nonimpaired Speakers,” by M. McClean, D. Beukelman, & K. Yorkston, *Journal of Speech, Language, and Hearing Research*, Volume 30, p. 279-280. Copyright 1987 by American Speech-Language-Hearing Association. Reprinted with permission.

4.4 Impaired strength and movement precision in swallowing

Given the contribution of both strength and movement precision impairments to motor functioning in speech and the limbs, the possibility of impaired coordination underlying dysphagia after stroke is an intriguing one. Although knowledge from largely voluntary behaviours such as speech and limb movement cannot be directly applied to swallowing behaviour, exploration of movement precision in swallowing is important as it reinforces the major shift in rehabilitation from peripheral strength-based exercises to modulation of centrally-mediated processes. It is possible that both weakness and a deficiency in skilled execution are present in dysphagia after neurological damage, albeit in varying levels. However, there are limited studies that have measured and compared the relative impact of strength and movement precision on swallowing and dysphagia.

Even though dysphagia is assumed to be predominantly characterised by weakness, the extent and mechanism by which weakness impacts swallowing remains unclear (Clark, 2003). Studies investigating aspects of muscle strength as it relates to swallowing and dysphagia have focused mainly on lingual strength. Air-filled bulbs placed on the surface of the tongue or adhered on the hard palate can be used to measure maximum isometric strength of the tongue pushing against the bulb in normal healthy participants and those who are highly skilled (e.g., trumpet players; Robin, Goel, Somodi, & Luschei, 1992), as well as patients with dysphagia (Robbins et al., 2007). Changes in lingual strength are not only associated with aging and sarcopenia (Clark & Solomon, 2012a; Robbins, Levine, Wood, Roecker, & Luschei, 1995; Tamine et al., 2010), but also can be a clinical indicator of impaired swallowing, as decreased lingual strength is correlated with symptoms of dysphagia during mealtimes (Yoshida et al., 2006). Tongue strength was found to be related to aspiration status, as healthy older adults who aspirated had significantly lower maximum and swallowing lingual strength than those who did not aspirate (Butler et al., 2011). A progressive lingual resistance exercise program has been shown to improve isometric lingual pressures, swallowing lingual pressures, and tongue volume in older adults (Robbins et al., 2005) and stroke patients with dysphagia (Juan et al., 2013; Robbins et al., 2007). These studies indicate that weakness of the tongue muscles may be a possible mechanism underlying post-stroke swallowing impairment.

However, the oral musculature is only one piece of the puzzle, and it is also important to understand how the strength of pharyngeal muscles can affect swallowing and dysphagia. A particularly crucial component of safe and efficient swallowing is the movement of the hyolaryngeal complex, achieved by contraction of the suprahyoid muscles. Since many of the submental muscles that elevate the hyoid are also involved in opening the jaw (mylohyoid, anterior belly of the digastric, and geniohyoid), strength of the suprahyoid muscles has been assessed by measuring jaw-opening force. Iida and colleagues (2013) measured the maximum jaw-opening force of healthy adults using a sthenometer, and found that both age and sex were significant factors in jaw-opening strength. Younger participants (aged below 70 years) had significantly greater jaw-opening force than older participants (over 70 years of age), while men had significantly greater jaw-opening force than women. However, jaw force in dysphagic versus healthy individuals has not been directly compared.

Similar to motor speech, swallowing is also a submaximal strength task, as demonstrated by the finding that healthy participants are able to generate maximum isometric pressure that is greater than pressure generated during swallowing (Robbins et al., 1995). Lingual pressure needed to produce safe and efficient swallowing is a proportion (approximately 55 – 65%) of the maximum pressure that the tongue can produce during isometric tasks (Todd, Lintzenich, & Butler, 2013). Healthy elderly participants have reduced maximum isometric pressure, but are able to maintain the same lingual pressure during swallowing as young participants (Nicosia et al., 2000; Robbins et al., 1995; Todd et al., 2013). This demonstrates that a reduction in maximum lingual pressure is part of healthy aging and does not affect the generation of adequate lingual pressure to swallow safely and effectively. Healthy elderly individuals also demonstrate adequate hyoid displacement during swallowing, despite having lower maximum isometric force than younger adults during jaw opening (Shinozaki et al., 2017).

Despite the limited amount of research on movement precision in swallowing, there are several lines of evidence that point towards the possibility of impaired movement precision causing dysphagia: 1) the cortex controls higher-level coordination and modulation of the swallowing response (Martin & Sessle, 1993; Mosier & Bereznaya, 2001), so damage to supramedullary structures might affect skilled motor execution; 2) skill training has demonstrated benefits in dysphagia rehabilitation (Athukorala et al., 2014), suggesting that it might be targeting underlying impairments in movement precision; 3) movement precision impairment has been demonstrated in the limb (Van Hedel et al., 2010; Wirth, Van Hedel, & Curt, 2008) and motor speech mechanisms (Duffy, 2005; McClean et al., 1987), and so this might also translate to the

swallowing system; and 4) swallowing is a submaximal task (Nicosia et al., 2000; Todd et al., 2013), so weakness cannot be the only component underlying dysphagia. Although there are other pathophysiological features likely contributing to dysphagia other than impaired strength and movement precision, the current assumption is that weakness is the predominant underlying mechanism of dysphagia. As a first step in challenging this assumption, research should focus on investigating whether impaired movement precision can be quantified, and differentiated from impaired strength.

Paik and colleagues (2008) measured strength and movement precision by comparing dysphagic stroke patients with dysphagic myopathy patients and healthy controls. By using frame-by-frame kinematic motion analysis of the hyoid bone on VFSS, the researchers were able to describe the movement trajectory of the hyoid bone and quantify hyoid velocity at each frame. These measures were used to reflect swallowing coordination. Maximum excursion of the hyoid bone in the horizontal and vertical planes was used as a measure of strength. The healthy group had a hyoid bone trajectory characterised by anterior-superior movement of the hyoid bone initially, a pause at the point of maximum excursion, and then posterior-inferior movement back to initial position. Stroke patients had similar extent of maximum anterior and superior excursion, but the pattern of movement was different. The hyoid bone did not demonstrate the normal pause, but instead, elevated during backwards movement before descending to the original position. The number of frames in which the hyoid bone velocity exceeded 2 cm/s (“speed peaks”) was also significantly higher in the stroke group. This suggests that the stroke group had relatively intact strength, but decreased accuracy in achieving a normal spatial and temporal pattern of movement. On the other hand, the extent of horizontal hyoid excursion and average hyoid velocity in myopathy patients was significantly reduced compared to stroke patients and healthy controls, reflecting underlying weakness. This study was insightful because it used a task that could analyse the separate contributions of movement precision and strength, and also reinforced the anatomical dissociation between movement precision and strength by comparing patients with dysphagia due to central versus peripheral damage. However, there were some limitations to this study. Hyoid bone displacement on VFSS can only infer suprahyoid muscle strength, and is easily confounded by other underlying pathophysiological impairments, such as reduced coordination or impaired tone. Use of jaw-opening force would be a better measure of suprahyoid muscle strength, as it directly measures isometric force of the suprahyoid muscles. In addition, kinematic motion analysis of VFSS is time-consuming and not easily applicable to clinical settings. Therefore,

future studies should be directed to developing simple, objective, and non-invasive measures of swallowing coordination and force that can be employed clinically.

Visuomotor tracking tasks, which tap into the ability to modulate timing and force, have been used to non-invasively measure movement precision of motor execution in swallowing impairment and for swallowing rehabilitation (Athukorala et al., 2014; Hands & Stepp, 2014; Malloy et al., 2014; Stepp et al., 2011). sEMG electrodes placed on the neck and submental region served as the input modality to control a cursor or waveform on the computer screen. Participants were asked to control the timing and force of their movement so that their response cursor was placed in a static or dynamic target on the screen, allowing for measurement of motor control and precision in healthy participants and patients with dysphagia.

Stepp et al. (2011) investigated the feasibility of anterior neck sEMG and a video game biofeedback interface to measure both movement precision and skill learning in a dysphagic brainstem stroke patient. An orange fish avatar was visualised on the left side of the laptop screen (Figure 4.3). Participants were instructed to move the fish vertically using any neck muscle activity (not specifically swallowing), with increased muscle activity leading to increased upward movement of the fish, and vice versa. Since the sEMG readings had large inter-participant variability secondary to anatomical differences, the presentation of biofeedback during the protocol was normalised to the maximum activity of effortful swallows completed at the start of each session. The goal of the task was for the participant's orange fish to move up and down to "eat" targets that move from the right to the left of the screen at a constant velocity. The targets were located at three different heights (33%, 66%, and 100% of the participant's maximum effortful swallowing amplitude) and also had three different lengths (corresponding to 2.8 s, 3.5 s, and 4.7 s of muscle activation). Participants completed seven trials per block, and ten blocks per visit.

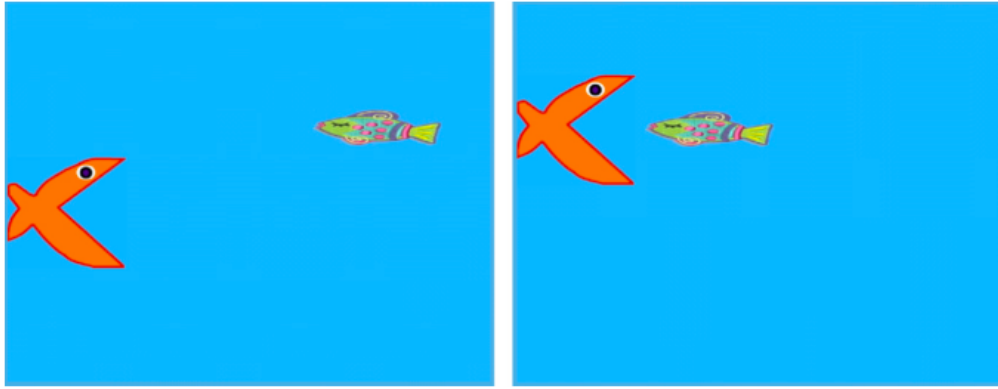


Figure 4.3. Two screenshots showing the swallowing target (small fish on right) which appears on the right side of the screen and moves towards the left at a constant velocity and vertical position. The participant controls the vertical position of the orange fish by controlling muscle activity of the neck, measured with sEMG. From “Feasibility of game-based electromyographic biofeedback for dysphagia rehabilitation,” by C. E. Stepp, D. Britton, et al., Proceedings of the 5th International IEEE/EMBS Conference on Neural Engineering, p. 234. © 2011 IEEE. Reprinted with permission.

Results show that the visuomotor task was able to identify movement precision impairment in the patient, as evidenced by the patient acquiring substantially fewer targets (mean of 0.9 per block) in the initial session compared to healthy individuals (mean of 3.3 targets). The authors concluded that since the healthy participants consistently performed better than the impaired patient, this measure of performance may be a reasonable indicator of voluntary control and coordination of neck muscle activity. Since the task’s targets were set at a percentage of the participant’s available muscle activity, even the patient with limited strength could participate, allowing for assessment of movement precision separately from strength level. However, since the participant was controlling the avatar with any available neck muscle activity (not just swallowing), research is needed to evaluate whether a similar biofeedback tool can be used to measure motor control and precision in swallowing. Another limitation of the study is the small sample size, which did not allow for statistical comparison between a healthy and disordered population.

4.5 Summary of evidence

There is currently an assumption that weakness is the predominant pathophysiological feature of dysphagia. Research has suggested that both impaired strength (decreased force generation) and movement precision (ability to voluntarily modulate swallowing force and timing in

performance of a complex, goal-oriented task) are mechanisms underlying dysphagia (Paik et al., 2008; Stepp et al., 2011). In the limb literature, the effects of strength and movement precision impairment on limb functioning have been elucidated by comparing the strength and movement precision performance of patients with central damage to patients with peripheral lesions, with the finding that patients of different aetiologies have distinct patterns of impairment (Van Hedel et al., 2010; Wirth et al., 2008). This leads to the question of whether a similar assessment can discriminate between strength and movement precision in swallowing. Development of an assessment tool that can objectively quantify and discriminate between swallowing strength and movement precision is important for improving diagnostic specificity.

A novel skill-training protocol using sEMG biofeedback presented submaximal targets on a screen, with a lower and upper boundary of 20 and 70% respectively of the individual's maximum muscle activity (Athukorala et al., 2014; Sella, 2012). Participants modulated amplitude of their submental muscle activity in order to meet the target. Previous research has demonstrated that normal swallowing requires approximately 42 – 47% of sEMG activity used during effortful swallowing (Huckabee et al., 2005; Wheeler-Hegland et al., 2008; Yeates et al., 2010). However, it is unknown whether individuals are able to volitionally use an even lower proportion of maximum swallowing muscle activity to meet submaximal targets as low as 20%, since no study has investigated the functional range of muscle activity used for swallowing in the healthy population.

The biomechanical impairments seen on VFSS are commonly used to diagnose underlying pathology in clinical practice, even though there are limited correlations between maximum isometric strength of submental muscles and corresponding biomechanical hyoid movement (Shinozaki et al., 2017). The relationship between impairments in movement precision assessed clinically and via instrumental methods has not been investigated, and requires further exploration.

Development of an assessment using non-invasive, objective, and clinical measures of strength and movement precision will be a valuable first step in understanding the underlying mechanisms of dysphagia after different neuromuscular lesions. Exploring the biomechanical correlates of these clinical measures will also provide further insight into strength and movement precision impairment. This first attempt at closing the knowledge gap regarding movement precision impairment will inform future research on improving diagnostic and treatment specificity.

PART B. EXPERIMENTAL STUDIES

Chapter 5. Objectives and Hypotheses

5.1 Range of submental sEMG activity during volitional swallowing: A methodological study (Study 1)

5.1.1 *Research questions*

What is the minimum and maximum peak amplitude and duration of submental sEMG activity that can be recruited for volitional swallowing, and where does regular swallowing lie along this continuum? Do age and bolus type affect the ability to modulate muscle activity?

5.1.2 *Objective*

To examine the extent to which healthy adults of different ages can alter magnitude of submental muscle activity, in order to determine the available range of sEMG activity for skill training sEMG target placement.

5.1.3 *Hypotheses*

Primary hypotheses

1. Normalised amplitude and duration of sEMG activity will be significantly decreased during minimum effort swallowing compared to regular effort swallowing.
2. Normalised amplitude and duration of sEMG activity will be significantly increased during maximum effort swallowing compared to regular effort swallowing.

Secondary hypotheses

3. There will be a significant interaction of age group and task, with older participants having a smaller increase in amplitude and duration of sEMG activity from regular to maximum effort tasks, and a smaller decrease in amplitude and duration of sEMG activity from regular to minimum effort tasks, compared to younger participants.
4. There will be a significant interaction of bolus type and task, with water swallows having a smaller increase in amplitude and duration of sEMG activity from regular to maximum effort tasks, and a smaller decrease in amplitude and duration of sEMG activity from regular to minimum effort tasks, compared to saliva swallows.

5.1.4 Rationale

The skill training protocol in the BiSSkiT software uses visual biofeedback targets placed on a computer screen at varying heights, between 20-70% of the individual's normalised sEMG amplitude during effortful swallowing (Athukorala et al., 2014; Sella, 2012). Participants were able to complete the task, however during pilot studies they anecdotally reported more difficulty acquiring skill training targets placed in the lower amplitude range. Individuals have the ability to increase amplitude and duration of muscle activity during maximum effort swallowing (Hind et al., 2001; Huckabee et al., 2005; Wheeler-Hegland et al., 2008; Yeates et al., 2010). It is unknown if, and to what degree, individuals are able to voluntarily decrease magnitude of muscle activity while still producing a swallowing response.

It is assumed that healthy individuals will maintain the ability to modulate muscle activity using minimal effort swallowing, thereby decreasing the magnitude and duration of muscle activity, however, this has not been directly assessed. Previous research revealed that older adults were less able to modulate oral pressures using effortful swallowing, compared to younger adults (Hind et al., 2001). The ability to decrease magnitude and duration of muscle contraction during minimum effort swallowing may also be restricted as people age. Finally, the effect of effortful swallowing can vary depending on the bolus being swallowed. Effortful saliva swallows produced a greater change in swallowing biomechanics than effortful water swallowing (Witte, Huckabee, Doeltgen, Gumbley, & Robb, 2008), therefore a similar interaction of task and bolus type may be expected with minimal effort swallowing.

5.1.5 Significance

This methodological study is important to refine methods for future research trials and clinical use of skill-based sEMG assessment and training protocols. These data will allow for accurate placement of the visual target used in skill training, such that the lower limit in particular is a meaningful and achievable target. In order to fully understand the effect of volitional effort on swallowing physiology, it is important to investigate the entire range of functional effort during muscle activation. If individuals are able to manipulate muscle activity in both directions, then this suggests that there is a minimum amount of muscle activity needed to generate functional swallowing, and that non-strength based protocols can be tailored to the entire functional range.

5.1.6 Proposed study

Forty healthy individuals (ten participants in each of four age groups: 20-39, 40-59, 60-79, 80+ years) will complete three swallowing conditions (maximum, regular, and minimum effort) with two bolus types (saliva and 5 mL water). Outcome measures will be peak amplitude during swallowing (normalised to maximum amplitude during effortful swallowing) and duration of swallowing onset to offset, measured using sEMG. Linear mixed effects models will be performed to estimate effects of swallowing task, and interactions of task with bolus and task with age.

5.2 Clinical classification of strength and movement-precision deficits in patients with dysphagia due to different aetiologies (Study 2)

5.2.1 Research question

Can a novel clinical assessment using objective measures of strength and movement precision discriminate between healthy controls and patients with dysphagia due to stroke versus myopathy?

5.2.2 Objective

To clinically classify healthy controls and patients with dysphagia due to stroke and myopathy into subgroups based on a novel, objective assessment of swallowing-related strength and movement precision.

5.2.3 Classification using diagnostic groups

5.2.3.1 Hypotheses

1. Patients with dysphagia associated with stroke will have significantly different performance on test measures of strength and movement precision compared to healthy controls. Specifically, patients with dysphagia associated with stroke will have:
 - a. Decreased effortful swallowing amplitude,
 - b. Decreased jaw-opening force,
 - c. Decreased swallowing hit rate,
 - d. Increased swallowing temporal peak-to-target error,
 - e. Increased swallowing amplitude peak-to-target error,
 - f. Decreased jaw-opening hit rate,
 - g. Increased jaw-opening temporal peak-to-target error, and

- h. Increased jaw-opening amplitude peak-to-target error.
- 2. Patients with dysphagia associated with myopathy will have significantly different performance on test measures of strength, but similar performance on test measures of movement precision, compared to healthy controls. Specifically, patients with dysphagia associated with myopathy will have:
 - a. Decreased effortful swallowing amplitude,
 - b. Decreased jaw-opening force,
 - c. No difference in swallowing hit rate,
 - d. No difference in swallowing temporal peak-to-target error,
 - e. No difference in swallowing amplitude peak-to-target error,
 - f. No difference in jaw-opening hit rate,
 - g. No difference in jaw-opening temporal peak-to-target error, and
 - h. No difference in jaw-opening amplitude peak-to-target error.

5.2.3.2 Rationale

A clinical strength and movement precision assessment has been developed to quantify submental strength (ability to generate force) and movement precision (spatiotemporal accuracy of movement) in swallowing. Since task performance may be different during voluntary and semi-reflexive tasks due to differences in neural control mechanisms (Doeltgen, Ridding, Dalrymple-Alford, & Huckabee, 2011), this study will explore measures of strength and movement precision during both jaw-opening and swallowing behaviours. Jaw-opening force, as assessed with a dynamometer, has been used as a measure of submental muscle force during swallowing, as these muscles are important for hyolaryngeal excursion during swallowing and also for opening the jaw (Hara et al., 2014; Iida et al., 2013). The effortful swallowing technique increases oropharyngeal pressures and submental muscle activity (Hiss & Huckabee, 2005), so measuring peak amplitude of sEMG activity during effortful swallowing may be a proxy measure of swallowing strength. During performance of a skill-based, goal-directed biofeedback task, number of acquired submaximal targets (“hit rate”) has been used as a reflection of swallowing precision (Athukorala et al., 2014; Sella, 2012). Quantifying temporal and amplitude errors between the response and the target would allow for greater specificity of measurement. In this novel assessment, movement precision will be measured using hit rate, temporal peak-to-target error, and amplitude peak-to-target error, during both swallowing and jaw-opening behaviours.

This study will explore the feasibility of this assessment based on the performance of three diagnostic groups: stroke patients with dysphagia, myopathy patients with dysphagia, and healthy controls. These patient diagnoses have been chosen because they are likely to have specific patterns of strength and movement precision abilities based on lesion location, and likely underlying pathophysiologic abnormalities. In the limb literature, patients with stroke-induced damage to cortical areas and descending pathways produce impairments in both strength and movement precision, while patients with peripheral damage (such as that seen in patients with myopathy) exhibit a characteristic presentation of weakness (Kitago & Krakauer, 2010; Van Hedel et al., 2010). In the swallowing musculature, impaired precision may be an underlying cause of dysphagia after central nervous system damage such as stroke (Paik et al., 2008; Stepp et al., 2011), while dysphagia in myopathy patients has primarily been characterised by weakness (Miller & Britton, 2011). Therefore, if this clinical test is sensitive to differential diagnosis of strength and movement precision, it would be expected that stroke patients in this study will have impaired performance on both the strength and movement precision measures on the novel clinical assessment, while myopathy patients with dysphagia will have impaired performance on strength test measures only.

5.2.4 Classification using cluster analysis

5.2.4.1 Hypotheses and rationale

Cluster analysis will also be used to explore whether there are subgroups within the participants based on strength and movement precision test performance. Given the exploratory nature of cluster analysis, which aims to uncover unknown patterns and groups in the data (James, Witten, Hastie, & Tibshirani, 2013), a priori hypotheses will not be formally posed for this analysis. Stroke patients have heterogeneous and varied dysphagia symptoms, with great inter-individual variability (Daniels et al., 2017, 1996; Perlman et al., 1994). Even though stroke patients as a group may have impaired strength and movement precision, there may be several different subtypes of dysphagia after stroke, each corresponding to a characteristic pattern of strength and movement precision impairment. This might be similar to how motor speech deficits after stroke can be classified into different dysarthrias based on patterns of impairment (Duffy, 2005). Healthy participants and patients with myopathy and dysphagia will be included in the cluster analysis as control groups, in order to internally validate the cluster solution and to aid in interpretation of the clinical assessment scores. Healthy participants are expected to have intact strength and movement precision, while myopathy patients were chosen to represent individuals with expected decreased strength and intact movement precision. If the

clinical assessment is sensitive to differentiating strength and movement precision in swallowing, healthy participants will likely be clustered together, and myopathy patients will be assigned to another cluster. Stroke patients might be spread out over several different clusters, with each cluster demonstrating different patterns of strength and movement precision impairment.

5.2.5 Significance

If the novel strength and movement precision assessment is able to differentiate between individuals with varying patterns of strength and movement precision, then this represents the first step in developing objective and quantifiable measurements of swallowing precision. The ability to identify deficits in spatiotemporal precision of movement would provide preliminary evidence to challenge the assumption that weakness is the predominant pathophysiology underlying dysphagia. Evidence of several dysphagia subtypes, even within the same diagnostic group, would suggest that different and specific rehabilitation approaches are needed for each pattern of impairment. Results from this study provide an initial framework for future research to investigate the pathophysiology of dysphagia and develop tailored assessment and treatment protocols.

5.2.6 Proposed study

Healthy controls (n=40) and patients with dysphagia due to stroke (n=60) and myopathy (n=20) will participate in a clinical assessment of strength and movement precision of the submental muscles during both swallowing and non-swallowing tasks, using sEMG, a biofeedback protocol, and dynamometry. Movement precision will be measured as the frequency that the peak of sEMG signal is placed in an on-screen target, and the peak-to-target error, during swallowing and jaw opening. Swallowing strength will be measured as peak sEMG amplitude during effortful swallowing, while non-swallowing strength will be measured as maximum isometric force during jaw opening. Performance between the three diagnostic groups will be compared. Cluster analysis will be used to explore the assignment of participants (regardless of diagnostic group) into clusters, based on test performance only.

5.3 Relationship between biomechanical measures of hyoid movement and physiological measures of strength and movement precision: An exploratory study (Study 3)

5.3.1 Research question

Do stroke patients with impaired strength performance and patients with impaired movement precision performance (as identified by the novel strength and precision assessment from Study 2) have different measures of biomechanics on VFSS?

5.3.2 Objective

The aim of this study is to explore the relationship between physiological measures of submental swallowing strength and movement precision, and kinematic measurements of hyoid movement.

5.3.3 Hypothesis

1. Patients identified as having only movement precision impairment in Study 2 will have significantly different measures of biomechanical movement on VFSS, compared to patients identified as having only strength impairment. Specifically, the two groups will demonstrate differences in:
 - a. Hyoid displacement,
 - b. Hyoid burst duration,
 - c. Hyoid velocity, and
 - d. Stage transition duration.

5.3.4 Rationale

In clinical practice, submental muscle function is inferred from visualising biomechanical assessment of hyoid bone displacement, duration, speed, and initiation on VFSS (Bingjie et al., 2010; Kim & McCullough, 2010; Paik et al., 2008; Sia, Carvajal, Lacy, Carnaby, & Crary, 2015). If the novel assessment of strength and movement precision in Study 2 can discriminate between stroke patients with predominantly impaired strength and those with predominantly impaired movement precision of the submental muscles during swallowing, one might expect to identify biomechanical measures of hyoid movement on VFSS that also differ between these patients. Since improvements in motor skill are associated with changes in limb trajectory and velocity (Shmuelof, Krakauer, & Mazzoni, 2012), this relationship between movement precision and kinematics might translate to swallowing behaviour. However, it is unknown whether the impaired movement precision group will have increased or decreased magnitude of biomechanical movement, compared to the impaired strength group. Given the novelty of the strength and movement precision test, and even the lack of an understanding of movement precision deficits in swallowing, it is not feasible to speculate on the direction of those differences.

5.3.5 Significance

Investigating the relationship between physiological and kinematic measures of impaired strength and precision will improve our understanding of the mechanisms underlying dysphagia. This information can be used for further development of the strength and movement precision assessment and its clinical applications. If clinical measures of strength and precision are correlated with biomechanical measures on VFSS, then this can provide some measure of external validation for using the new assessment. If there are limited or no significant associations between the strength and precision assessment and VFSS measures of biomechanics, then this suggests that there may be other factors influencing hyoid kinematics, in addition to movement force and precision.

5.3.6 Proposed study

Eight stroke patients will be identified from Study 2, who demonstrate impairments in measures of strength only (n=4), or impairments in measures of movement precision only (n=4). Impairment will be defined as performance poorer than the 95% confidence interval of the healthy group's mean. The patients will participate in a VFSS examining four aspects of hyoid kinematics (maximum displacement, duration of hyoid burst, velocity of hyoid movement, and stage transition duration) during swallowing of 5 mL thin liquid and 5 mL puree. VFSS measurements will be compared between the two groups.

Chapter 6. Range of submental sEMG activity during volitional swallowing: A methodological study (Study 1)

6.1 Introduction

Skill-based protocols using sEMG biofeedback have recently emerged as a promising tool for swallowing rehabilitation (Athukorala et al., 2014; Stepp et al., 2011). Patients voluntarily increase and decrease the precision (accuracy of force and timing) of muscle activity during swallowing to control a sEMG signal displayed on a screen in real-time. Relative amplitude of muscle activity is represented by vertical movement of the signal. The screen height is calibrated to the participant's maximum sEMG activity during effortful swallowing. A key aspect of these protocols is measuring movement precision of swallowing behaviour to meet the amplitude and temporal requirements of an on-screen target.

Rather than repetitive practice of a single task, it has been found that introducing task variability can promote improved skill learning (Krakauer, 2006). To this end, skill-based training protocols in dysphagia have been designed so that the screen position of the target changes between trials (Athukorala et al., 2014; Sella, 2012; Stepp et al., 2011). For example, in one skill-training protocol using anterior neck sEMG biofeedback, targets were displayed at different heights on the screen (33%, 66%, and 100% of the participant's maximum effortful swallowing amplitude; Stepp et al., 2011). Participants were required to modulate relative amplitude of muscle contractions to meet the target. In another protocol, a square target moved to a random horizontal and vertical location at every trial, but did not move higher than 70%, or lower than 20%, of the participant's maximal voluntary contraction (Athukorala et al., 2014; Sella, 2012). The upper ceiling of 70% was chosen so that only submaximal amplitude targets were displayed, and the task would not be confounded by possible strength training effects. Exercise physiology research in the limb literature has demonstrated that increases in strength occur when training at intensities of 60%, or higher, of the maximum force generated in a single repetition (Burkhead et al., 2007; Porter, 2000). Strength training protocols in dysphagia have used training targets set at 60-80% of an individual's maximum force (Kim & Sapienza, 2005; Robbins et al., 2007), while skill training protocols have used submaximal targets set below 70% (Athukorala et al., 2014; Sella, 2012). However, the lower border of 20% has not been justified. Anecdotal evidence from pilot studies revealed that participants had more difficulty controlling their swallowing amplitude to meet targets near the lower portion of the screen. Regular swallowing uses approximately 45% of the muscle activity required during effortful swallowing (Huckabee et al., 2005; Wheeler-Hegland et al., 2008). It is unknown if individuals

are able to reduce muscle activity below normal, and whether the lowermost range of 20% of maximal muscle activity is physiologically achievable, as there may be a minimum magnitude of muscle activity needed for functional swallowing.

The ability to modulate amplitude and temporal aspects of swallowing using volitional effort is central to behavioural rehabilitation techniques such as effortful swallowing (Clark & Shelton, 2014) and the Mendelsohn manoeuvre (McCullough & Kim, 2013). It has been previously documented that volitionally increasing effort during swallowing is associated with an increase in sEMG amplitude (Doeltgen, Ong, Scholten, Cock, & Omari, 2017; Huckabee et al., 2005; Wheeler-Hegland et al., 2009). Increased effort during swallowing also prolongs swallowing duration, as measured with sEMG (Ding et al., 2003), VFSS (Hind et al., 2001; Molfenter, Hsu, & Lazarus, 2017), and manometry (Hiss & Huckabee, 2005; Witte et al., 2008). However, the effect of volitionally reducing effort on amplitude and duration of sEMG muscle activity during swallowing has not been investigated.

The difference between the maximum capacity of a muscle and the submaximal capacity needed during functional swallowing has been termed functional reserve (Ney, Weiss, Kind, & Robbins, 2009). There is evidence of declining functional reserve in the healthy elderly, where older individuals have a significantly reduced maximum isometric lingual pressure compared to younger adults, but lingual pressure during swallowing is preserved (Nicosia et al., 2000; Robbins et al., 1995). These healthy age-related changes in *functional* reserve (that is, difference in muscle capacity between maximum voluntary contraction and regular swallowing) suggests that there may be similar effects on *swallowing* reserve, defined as the difference between muscle contraction during maximum effort swallowing and regular effort swallowing (Yeates et al., 2010). However, research in this area has demonstrated inconsistent results. In one study comparing submental sEMG amplitude during regular and effortful swallowing, both younger and older adults had significantly increased sEMG amplitude during effortful swallowing (Yeates et al., 2010). There was no difference in the magnitude of swallowing reserve between the younger and older groups, suggesting that age did not impact regular and effortful swallowing. Another study measuring swallowing reserve found that both younger and older adults had increased oral pressure during effortful swallowing. However, the difference between regular and effortful swallowing was greater in the younger group (Hind et al., 2001). Therefore, these studies suggest that there may be an influence of age on swallowing reserve, where the size of the reserve may be greater in younger compared to older adults. Additionally, submental sEMG activity can be influenced by the type of bolus ingested.

Water swallows had a higher sEMG signal value than saliva swallows (Gupta, Reddy, & Canilang, 1996; Vaiman, Eviatar, & Segal, 2004). Bolus type may also affect swallowing reserve. Effortful swallows performed with saliva had a significantly greater effect on lowering nadir UES pressures than effortful swallows performed with a water bolus (Witte et al., 2008). These differences demonstrate the need to take age and bolus type into account when investigating sEMG activity and volitional effort. If there is a reserve between regular and maximum effort swallowing, there may be a similar difference between regular and minimum effort swallowing, which is also influenced by age and bolus.

The primary goal of this methodological study was to investigate the extent to which healthy adults are able to modulate muscle activity using minimum and maximum volitional effort, thus identifying the lower limit of available muscle activity for skill training target placement. Results will also answer whether there might be a difference between regular and minimum effort swallowing, similar to that between regular and maximum effort swallowing. Evidence of this difference is important because it would suggest that a minimum threshold needs to be surpassed in order to generate a physiological, patterned swallowing response in healthy individuals. This study will also examine the influence of age and bolus type on muscle activity during both maximum and minimum effort swallowing tasks. These findings would hold significance for future research and clinical work that may incorporate different boluses into skill training protocols, with patients across the lifespan.

6.2 Methodology

6.2.1 *Participants*

Participants were recruited via written and verbal advertisement. Forty-three healthy adults (22 female, 21 male) participated in the study, representing four age groups: 20-39 years (n=10), 40-59 years (n=11), 60-79 years (n=12), and 80+ years old (n=10). Gender was approximately matched within and across age groups. All participants reported a negative history for neurological or swallowing impairments. Written informed consent was obtained prior to data collection, and the study was approved by the appropriate regional Human Ethics Committee (see Appendices).

6.2.2 *Experimental procedure*

Participants were seated comfortably during the study. sEMG data were collected using the KayPentax Digital Swallowing Workstation (KayPentax, Lincoln Park, NJ, USA) and self-

adhesive triode patch electrodes. Reducing the skin-electrode impedance prior to electrode placement is important for improving the quality of the sEMG signal (Merletti & Parker, 2004). The common method of wiping the skin with alcohol only reduces impedance by approximately 40%, while the practice of “peeling,” or repetitive application and removal of adhesive tape on the skin, has been found to reduce impedance by over 70% (Merletti & Parker, 2004). In this study, skin preparation included shaving the skin if there was hair, “peeling” of the skin surface under the chin using adhesive tape, and cleaning of the skin using an alcohol wipe (Stepp, 2012). The submental muscle group was identified via palpation between the mental symphysis anteriorly and the superior palpable border of the thyroid posteriorly. The two recording electrodes were placed at midline over the submental muscle group in the anterior-posterior plane, with the ground electrode oriented laterally. Raw sEMG signals were sampled at 250 Hz, bandpass filtered (50 – 250 Hz), integrated (50 ms time constant), and rectified.

Participants completed six different swallowing conditions: 1) maximum effort saliva swallow, 2) maximum effort water swallow, 3) regular effort saliva swallow, 4) regular effort water swallow, 5) minimum effort saliva swallow, and 6) minimum effort water swallow. Instructions for a regular effort swallow were: “Swallow like you normally would.” Instructions for a “hard swallow,” or maximum effort swallow, were: “As you swallow, swallow hard with all the muscles in your mouth and throat.” For a “soft swallow,” or minimum effort swallow, instructions were: “Swallow as lightly as you can, with as little effort as possible.” Water boluses were self-presented using a Provale™ cup (Reliant Medical Products, Birmingham, AL, USA), which dispensed a fixed amount (5 mL) of water for every trial.

The researcher trained the participant on execution of the tasks prior to data collection. During training only, participants were able to view the sEMG signal in real time on the computer screen for visual biofeedback. The researcher encouraged them to maximise the signal peak amplitude during effortful swallowing and minimise the amplitude during minimum effort swallowing. Participants practiced each of the three swallowing tasks at least three times, or until adequate comprehension and execution of the task could be demonstrated. During data collection, each of the six conditions were repeated five times for a total of 30 trials. The 30 trials were completed in randomized order at a rate of approximately one swallow every 30 seconds. Prior to each trial, participants were instructed on which of the six swallowing conditions to perform, and then given a verbal command to swallow 2-3 seconds later. For water swallows, participants were instructed to sip the fixed amount of water from the cup,

bring their head and neck back to neutral position, and hold the water in the oral cavity for a few seconds. A stable sEMG baseline was ensured before giving the participant the verbal command to swallow now. Participants were instructed to ingest the 5 mL bolus in a single swallow. No verbal or visual biofeedback of performance was provided during data collection.

6.2.3 Outcome measures

Each swallow was marked during data collection by the researcher, using the tagging function of the DSW. The outcome measures extracted from each swallowing waveform were sEMG amplitude (μV) and swallowing duration (s). Amplitude was extracted by manually selecting and zooming in on the waveform segment associated with the swallowing event; the software then extracted the maximum amplitude within the selected segment. Raw amplitude values were normalised relative to the maximum amplitude value from the patient's five effortful swallows, which was assigned a value of 100%. Since there can be large variability in raw sEMG amplitudes between participants due to inter- and intra-individual differences (e.g., in skin fold thickness, muscle activity, and electrode contact), normalising the peak amplitude data allows for between-participant comparisons.

Previous studies have defined swallowing duration as the onset of a dramatic or sharp increase of the sEMG signal from a baseline resulting in the peak amplitude, to the point where the waveform returns to a similar baseline amplitude level (Crary et al., 2006; Crary, Carnaby, & Groher, 2007; Ercolin et al., 2013; Perlman et al., 1999). In order to further quantify this measure, the onset of a dramatic increase in the sEMG signal from baseline was defined in this study as a greater than 45 degree slope in a 10 s time window.

A total of 1290 swallows (30 swallows from each of 43 participants) were measured. Five percent (65/1290) of the total measured swallows were discarded from data analysis as there was no steep increase in the waveform greater than 45 degrees, or if it was impossible to distinguish the swallowing peak from extraneous muscle activity. Seventeen percent (11/65) of the discarded swallows were maximum effort swallows, 34% (22/65) were regular effort, and 49% (32/65) were minimum effort swallows.

Visual inspection of residual plots for duration data revealed no obvious deviations from homoscedasticity or normality. However, peak amplitude data deviated from homoscedasticity and normality, and were subsequently natural log transformed to meet these assumptions.

6.2.4 Statistical analysis

Statistical analyses of the data were completed using RStudio software, version 3.2.4. Inter-rater reliability between the primary rater and a secondary rater with expertise in sEMG measurement of swallowing was calculated on a random 20% of the dataset for swallowing duration, using intraclass correlation coefficients. Linear mixed effect analyses were completed to investigate the relationship between swallowing task, age, and bolus type on outcome measures. Fixed effects entered in the model were swallowing task (maximum, regular, minimum effort swallowing), age group (20-39, 40-59, 60-79, 80+) and bolus type (water, saliva). Random intercepts for participant and by-participant random slopes for the effect of task were included to control for individual differences, while random intercepts for the five replications of each swallowing condition were included to control for any possible trial effect. Interactions of task x bolus type and task x age group were analysed, as well as the main effects of task, age group and bolus. If significant interactions or main effects were found, post-hoc analyses were completed using pairwise comparisons to determine significant differences between levels of effort, using Tukey adjustments to correct for Type 1 error. All analyses were completed separately for the two outcome measures of amplitude and duration. Statistical significance was assessed by comparing the full model against a model without the effect in question, using likelihood ratio tests with an alpha level of .05.

6.3 Results

High inter-rater reliability was found for measuring swallowing duration, ICC (1, k) = .82, 95% CI [.77 - .85]. Table 6.1 displays descriptive statistics for sEMG amplitude and duration, displayed by task, bolus type, and age group.

Table 6.1

Observed Sample Means and Standard Deviations for sEMG Peak Amplitude and Duration by Task, Bolus, and Age

Task	Bolus	Age group	Peak amplitude (%)	Duration (s)
			Mean \pm SD	Mean \pm SD
Maximum	Saliva	20-39	81.1 \pm 14.1	1.34 \pm 0.40
		40-59	75.2 \pm 17.6	1.21 \pm 0.31
		60-79	79.8 \pm 17.7	1.36 \pm 0.49
		80+	80.6 \pm 15.6	1.33 \pm 0.42
	Water	20-39	79.7 \pm 15.3	1.32 \pm 0.36
		40-59	81.8 \pm 16.2	1.18 \pm 0.27
		60-79	84.1 \pm 15.3	1.18 \pm 0.34
		80+	84.1 \pm 15.9	1.23 \pm 0.33
Regular	Saliva	20-39	34.5 \pm 14.1	0.92 \pm 0.24
		40-59	30.3 \pm 12.3	0.98 \pm 0.25
		60-79	41.5 \pm 17.8	1.03 \pm 0.29
		80+	60.1 \pm 28.4	1.12 \pm 0.41
	Water	20-39	36.6 \pm 17.1	0.95 \pm 0.33
		40-59	34.8 \pm 18.7	0.94 \pm 0.22
		60-79	43.3 \pm 22.0	0.91 \pm 0.28
		80+	47.5 \pm 22.0	0.96 \pm 0.29
Minimum	Saliva	20-39	24.3 \pm 13.4	0.87 \pm 0.27
		40-59	22.6 \pm 10.4	0.88 \pm 0.22
		60-79	29.3 \pm 10.6	0.99 \pm 0.34
		80+	41.7 \pm 21.8	0.99 \pm 0.35
	Water	20-39	30.2 \pm 15.6	0.94 \pm 0.35
		40-59	28.1 \pm 18.4	0.93 \pm 0.30
		60-79	36.7 \pm 20.2	0.90 \pm 0.31
		80+	37.6 \pm 20.2	0.88 \pm 0.27

Note. Peak amplitude expressed as a percentage of the maximum value acquired during five maximum effort swallows.

6.3.1 Effect of task

Regardless of age or bolus type, there was a main effect of task on sEMG amplitude [$\chi^2(2) = 72.27, p < .001$] and duration [$\chi^2(2) = 23.60, p < .01$]. As seen in Figure 6.1, peak amplitude of maximum effort swallows ($M = 80.8\%$, 95% CI [79.3, 82.4]) was significantly higher than regular effort swallows ($M = 40.9\%$, 95% CI [38.8, 43.0]; $p < .001$). Amplitude for minimum effort swallows ($M = 31.1\%$, 95% CI [29.4, 32.9]) was significantly lower than regular effort swallows ($p < .001$). Figure 6.2 demonstrates that duration of maximum effort swallows ($M = 1.27$ s, 95% CI [1.23, 1.30]) was significantly longer than regular swallows ($M = .97$ s, 95% CI [.95, 1.00]; $p < .001$), but there was no difference in duration between regular and minimum effort swallows ($M = .92$ s, 95% CI [.89, .95]; $p = .06$).

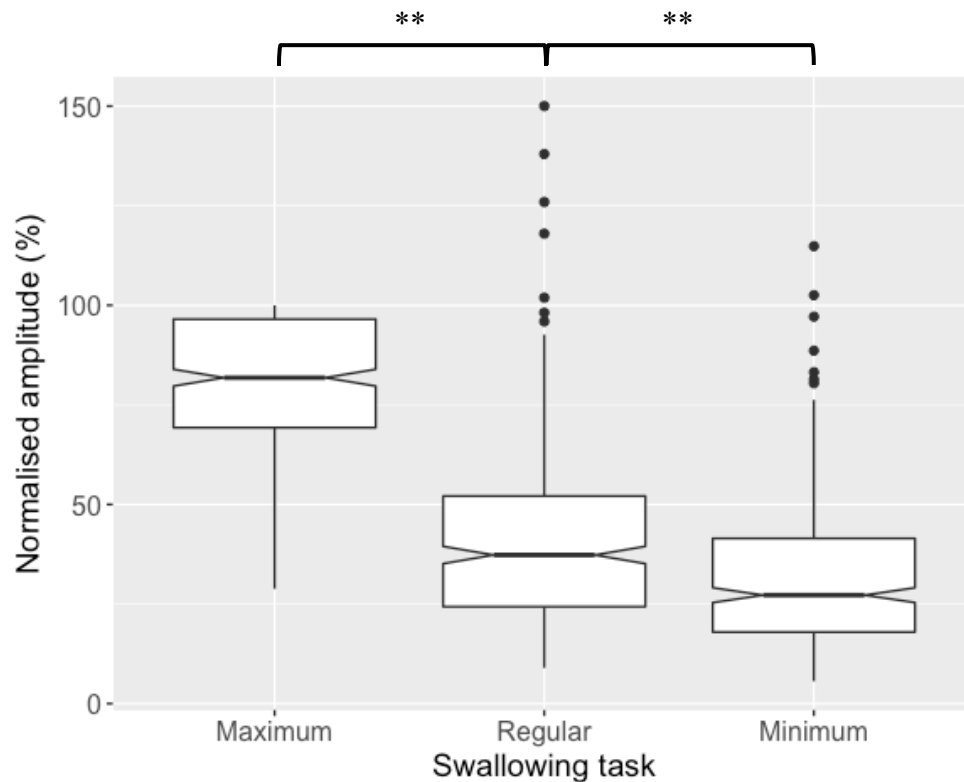


Figure 6.1. Boxplots of the normalised peak amplitude by swallowing task. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical

whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval. $**p < .01$.

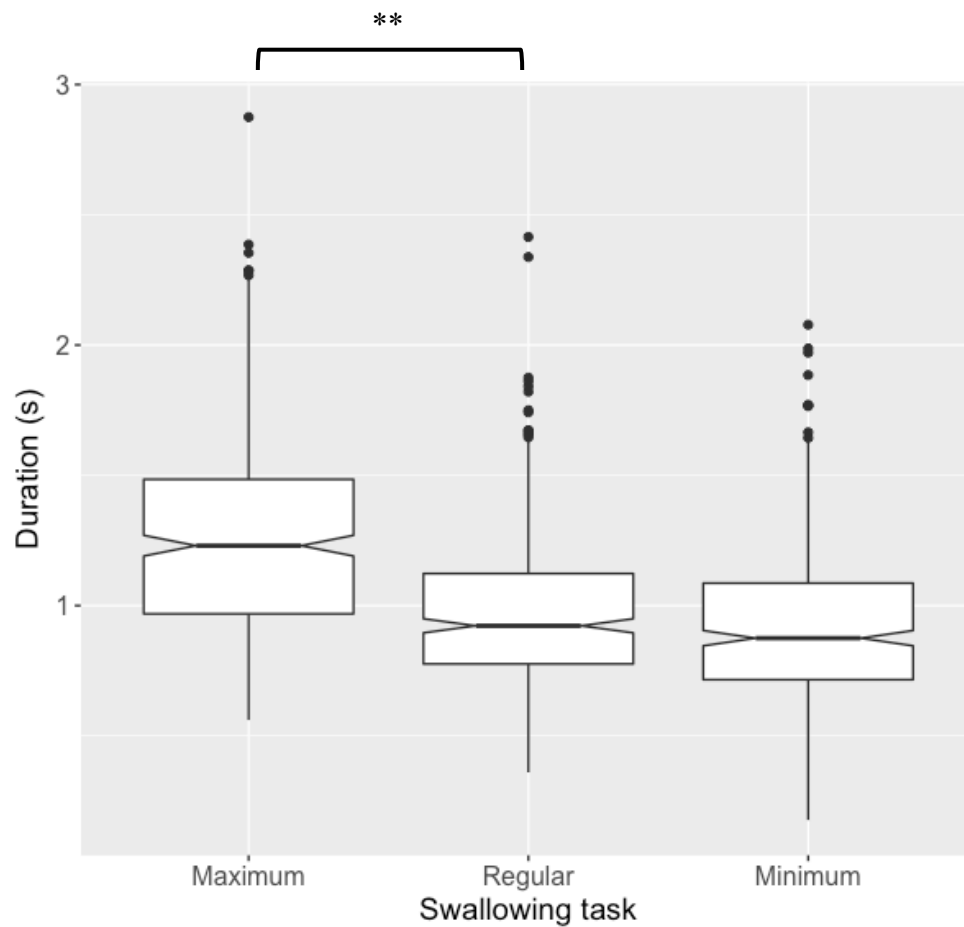


Figure 6.2. Boxplots of swallowing duration by task. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval. $**p < .01$.

6.3.2 Relationship between task and bolus

As shown in Figure 6.3, there was no significant interaction of task and bolus type for sEMG amplitude [$\chi^2(2) = 5.84, p = .054$]. Similarly, there was no significant interaction of task and bolus type noted for swallowing duration [$\chi^2(2) = 5.41, p = .07$], as presented in Figure 6.4. A main effect of bolus on amplitude [$\chi^2(1) = 5.23, p = .03$] and duration [$\chi^2(1) = 15.92, p < .001$] was found, with water swallows having significantly higher amplitude and shorter duration than saliva swallows across all levels of the swallowing tasks.

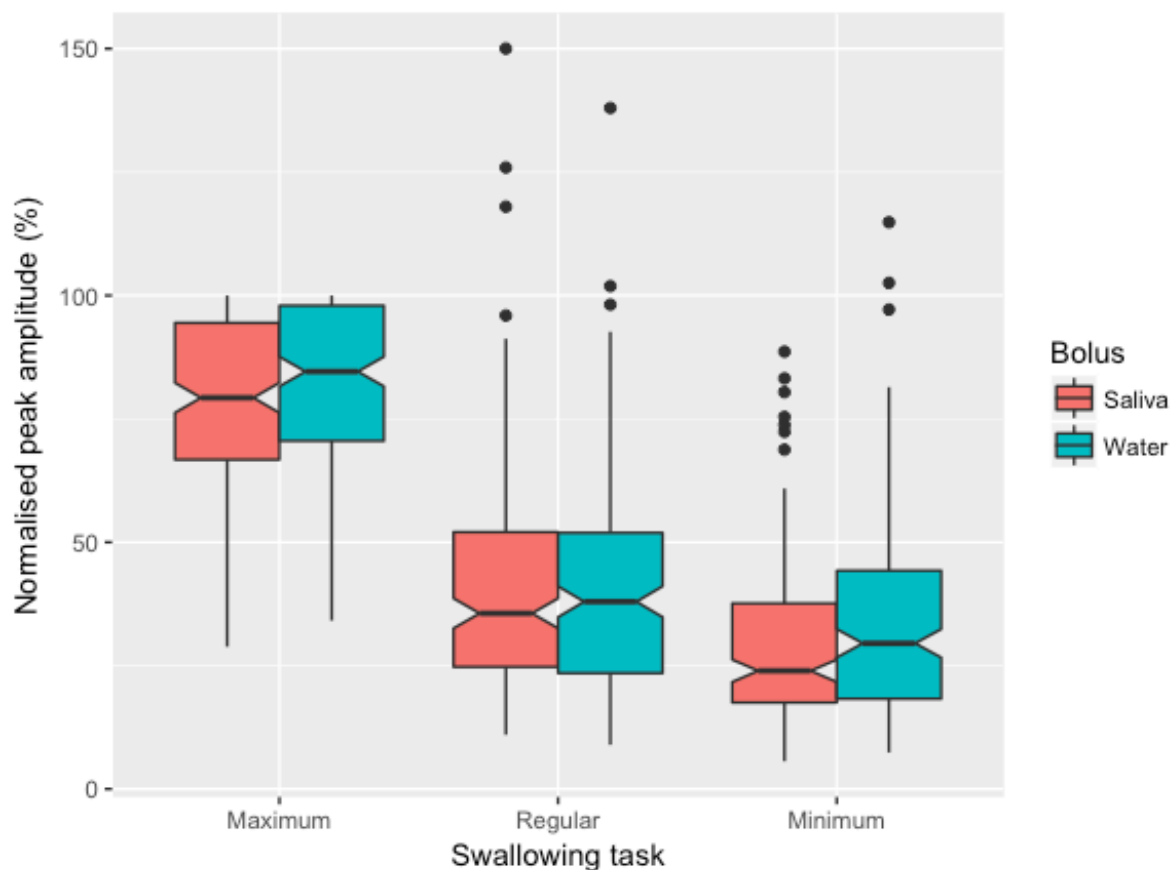


Figure 6.3. Boxplot of the normalised peak amplitude by swallowing task and bolus type. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval.

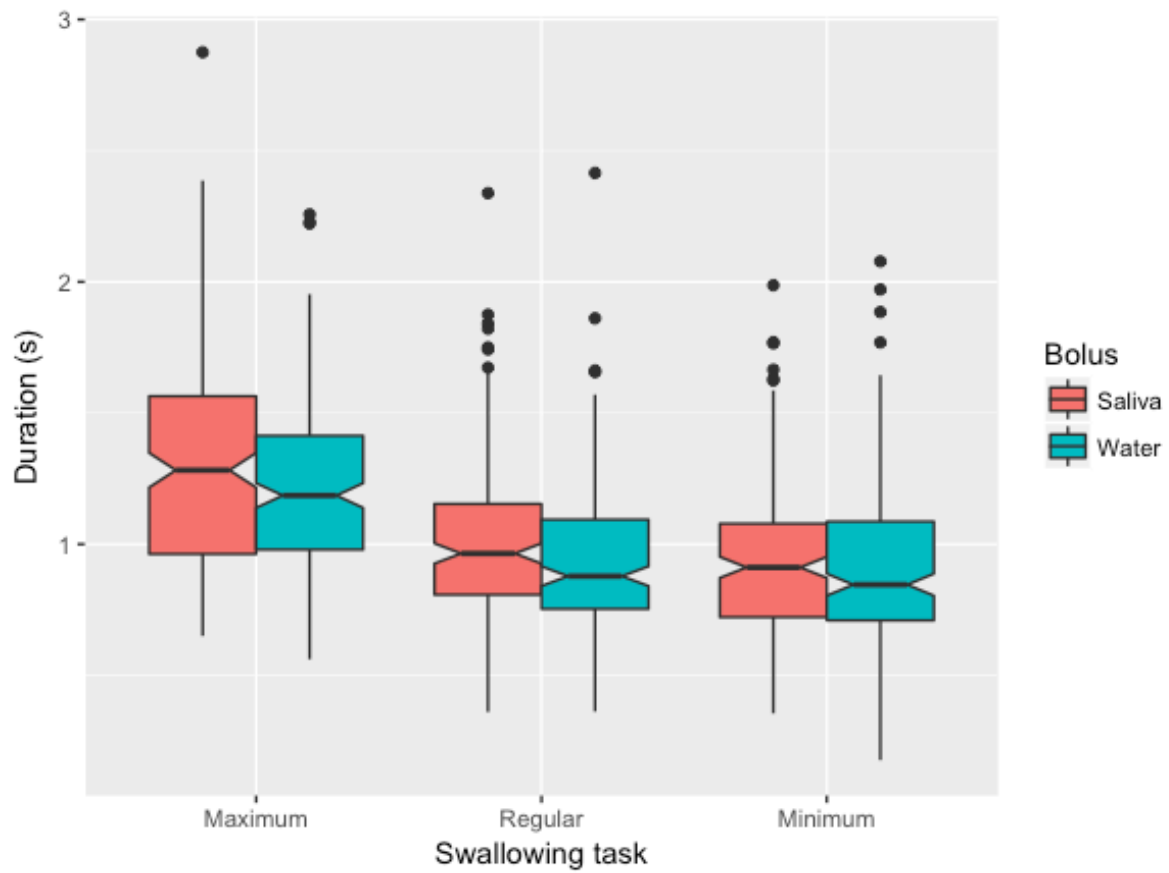


Figure 6.4. Boxplot of the normalised peak amplitude by swallowing task and bolus type. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval.

6.3.3 Relationship between task and age group

There was no significant interaction of task and age group [$\chi^2(6) = 6.67, p = .35$] nor main effect of age group [$\chi^2(3) = 2.13, p = .55$] on sEMG amplitude (Figure 6.5). Likewise, there was no significant interaction of task and age group [$\chi^2(6) = 8.32, p = .22$] nor effect of age group [$\chi^2(3) = 0.51, p = .92$] on swallowing duration (Figure 6.6).

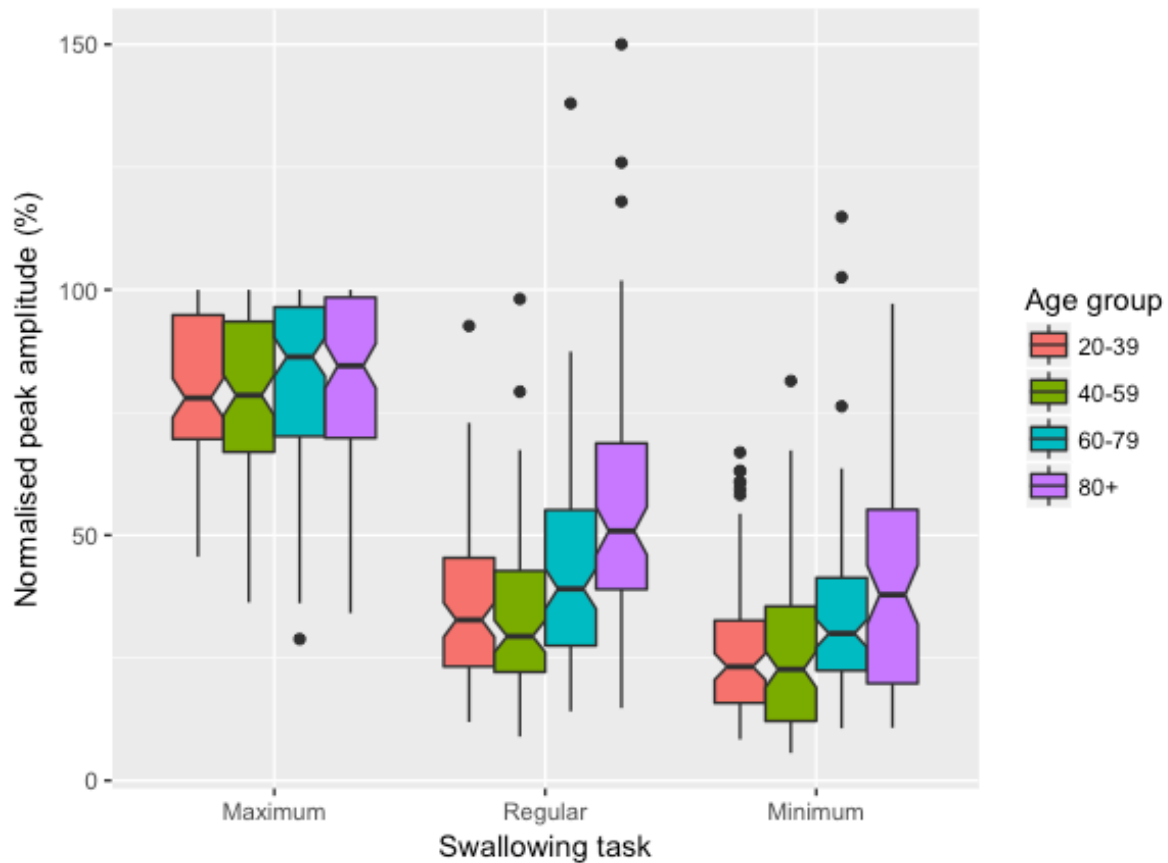


Figure 6.5. Boxplot of the normalised peak amplitude by swallowing task and age group. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval.

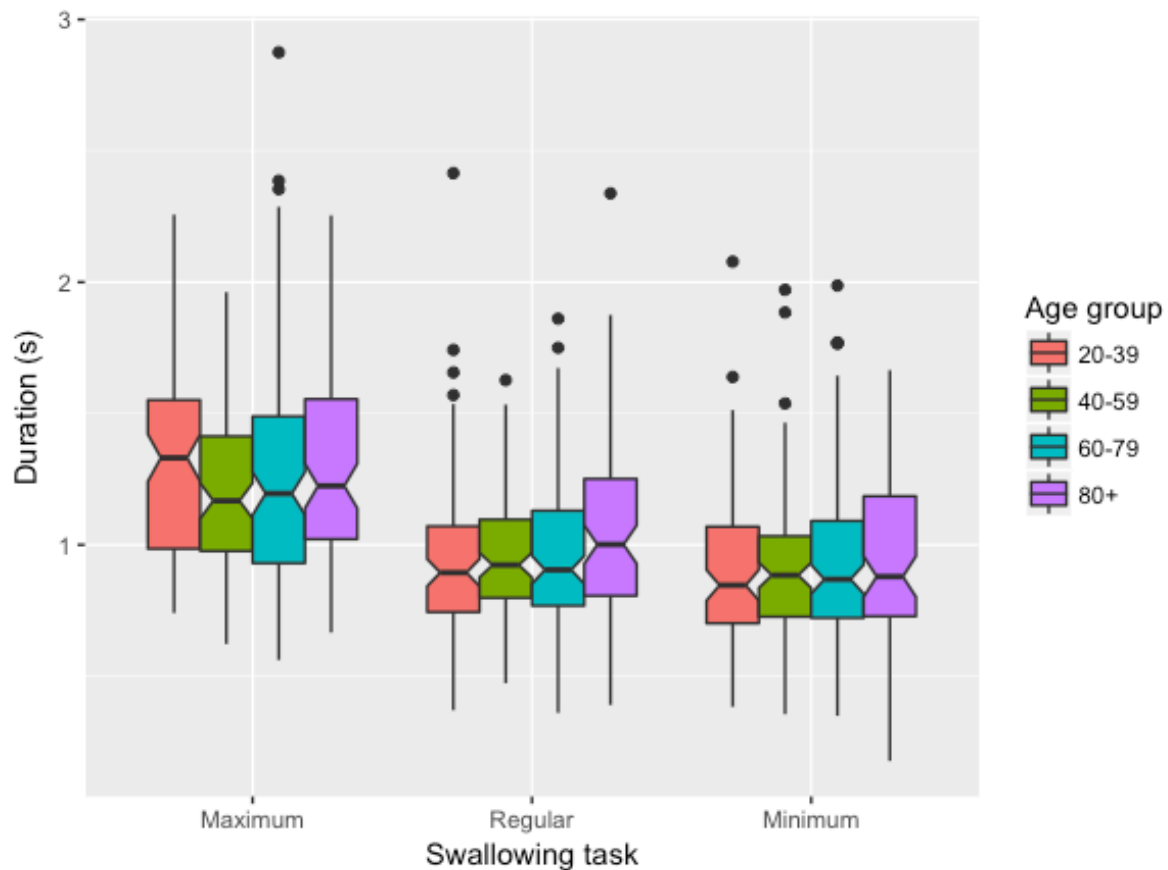


Figure 6.6. Boxplot of the normalised peak amplitude by swallowing task and age group. Horizontal lines indicate lower and upper quartiles of the data, centre line denotes the median, and vertical whiskers represent 1.5 times the interquartile range. Notches around the median mark the 95% confidence interval.

6.4 Discussion

The main goal of this study was to identify the minimum to maximum normalised amplitude range of muscle activity used during swallowing in healthy adults, so that appropriate positioning of skill-training targets in the lower range could be determined for use in the subsequent clinical study. Results show that healthy adults are able to volitionally decrease swallowing amplitude in order to acquire submaximal targets, but the 95% confidence interval for the minimum peak sEMG activity needed to generate functional swallowing was 29 – 34%. Therefore, skill-training targets should not be set lower than approximately 30% of the patient's maximum muscle activity during effortful swallowing for healthy participants. By incorporating this lowermost limit, individuals should have improved participation in skill training protocols because the task is physiologically achievable.

The finding that maximum effort swallowing significantly increased amplitude and duration of muscle activity from regular swallowing provides support for the concept of a swallowing reserve, reported in previous studies (Yeates et al., 2010). The relatively small proportion of submental sEMG amplitude needed for regular swallowing compared to effortful swallowing was found to be consistent with other reports in the literature (Doeltgen et al., 2017; Huckabee et al., 2005; Wheeler-Hegland et al., 2008; Yeates et al., 2010). A difference in sEMG amplitude was also found between regular and minimum effort swallowing, although this difference was smaller than that between regular and maximum effort swallowing. This suggests that regular swallowing is more similar physiologically to minimum effort swallowing, reinforcing the concept that swallowing is a submaximal task (Nicosia et al., 2000). These findings conceptually challenge the heavy emphasis on maximum-effort muscle strengthening as a rehabilitation approach, as only a proportionally small amount of available muscle activity is needed for regular, functional swallowing. Rehabilitation techniques that emphasise progressive strengthening at high power levels may not be appropriate for all patients. Given these results, training the precision and accuracy of movement at a submaximal level may be more logical than increasing maximum strength.

Although the sEMG amplitude of minimum effort swallowing was significantly different from regular effort swallowing, swallowing duration was not. The rapid sequencing of certain swallowing events is necessary for airway protection, and the short duration of swallowing may already be optimised for efficiency and safety. Even though there is variability in temporal measures of swallowing within and between healthy individuals, the range of values is still under 1 second. A systematic review of 46 studies found that mean UES opening duration ranged from .21 to .67 s, and mean laryngeal closure-to-UES opening interval ranged from -.16 to .02 s (Molfenter & Steele, 2012). This may explain why swallowing duration could not be significantly shortened with minimal effort swallowing.

The influence of age and bolus on the ability to modulate sEMG amplitude and duration using volitional effort was investigated. It was hypothesised that age would affect the ability and extent to which submental sEMG activity could be increased and decreased. Results of this study did not support this hypothesis. Older participants were able to maintain the same magnitude and duration of muscle activity during regular swallowing as younger participants, and were able to increase and decrease muscle activity during maximum and minimum effort swallowing respectively, to the same extent as younger people. This reinforces the idea that elderly people preserve the ability to alter submental sEMG amplitude and lingual pressure

using effortful swallowing, compared to regular swallowing (Yeates et al., 2010). This ability is maintained despite the possibility of age-related deterioration in isometric strength due to sarcopenia (Ney et al., 2009). Since possible reductions in isometric strength did not negatively impact functional swallowing in the healthy elderly in this study, this raises the question whether a large functional reserve is necessary for safe and efficient swallowing. Rehabilitation for impaired swallowing might be more effective if it focused on re-establishing the flexibility of the swallowing response, instead of the maximum force of muscle contraction.

It was hypothesised that differences seen in sEMG muscle activity due to volitional effort would be smaller when swallowing water compared to saliva. This hypothesis was not supported, as the effect of volitional effort on sEMG muscle activity remained constant regardless of water or saliva swallowing. Results from previous research have been inconclusive on this matter using alternative outcomes, with effortful and noneffortful swallowing demonstrating the same effect on oropharyngeal and midpharyngeal pressure in both water and saliva swallows, but significantly different effects on UES pressure (Witte et al., 2008). Another finding in this study was of increased amplitude and shortened duration of muscle contraction during water swallows, regardless of task effort. This was consistent with previous research showing that water swallows have a shorter duration of submental sEMG and tongue pressure activity (Perlman et al., 1999; Witte et al., 2008) and higher amplitude of muscle activity (Gupta et al., 1996) compared to saliva swallows. While skill training is usually completed with saliva swallowing, incorporating bolus swallows into therapy may increase the specificity of the task (Crary, Carnaby-Mann, Groher, & Helseth, 2004). Results from this study suggest that volitional modulation of muscle activity is still possible when swallowing with a bolus, although the placement of the visual target may need to be adjusted to account for the higher amplitude and shorter duration of water swallows.

There have only been two previously published studies that have documented the biomechanics of minimal effort swallowing (Garcia et al., 2004; Huckabee et al., 2014). Both reports differed from the current study in that they involved patients with dysphagia, and used different outcome measures (pharyngeal pressures using manometry, Huckabee et al., 2014; and timing and displacement of structural movement using VFSS, Garcia et al., 2004). In a case study on a patient with dysphagia after removal of a brainstem tumour, it was suspected that maladaptive behaviour had developed after using effortful swallowing as a compensatory technique for 5 months (Garcia et al., 2004). The patient demonstrated improved swallowing by using a more relaxed, “effortless” swallow. Perhaps the reduced muscle contraction generated during

minimal effort swallowing improved swallowing by “normalising” maladaptive behaviours caused by the use of effortful swallowing. Interestingly, both the above studies demonstrated that minimal effort swallowing resulted in a prolongation or later onset of swallowing events, while this study found a nonsignificant reduction in swallowing duration. This could be explained by the different populations, assessment techniques, and outcome measures studied.

Limitations of this study include the lack of instructions given to participants on tongue strategy during effortful swallowing. Instructions to emphasise tongue-to-palate pressure during effortful swallowing was found to increase submental sEMG amplitude and oropharyngeal pressure more than when told to inhibit tongue-to-palate pressure (Huckabee & Steele, 2006). This study did not explicitly instruct participants on which strategy to use, which may explain why a small number of participants had regular and minimum effort swallows with a higher amplitude than their maximum effort swallows. Another limitation was the subjective determination of sEMG onset and offset to calculate swallowing duration. Trials with a water bolus demonstrated a higher baseline amplitude prior to swallowing due to oral holding of the bolus, compared to saliva swallows with no pre-swallow hold. This higher pre-swallow baseline may have affected accurate identification of swallowing onset and thus calculation of the swallowing duration measure. Identification of swallowing onset in minimum effort trials was also challenging because the swallowing peak was not as obvious, and the sEMG onset slope resulting in the peak was less steep than those seen in regular and maximum effort swallows. This resulted in almost half of the discarded swallows being minimum effort trials. While there was high inter-rater reliability of the swallowing duration measure in this study, using computer software to mark onset and offset in an objective manner would improve the validity of measurement. Finally, the sampling rate for acquiring sEMG data was likely set too low; future studies should use a rate of at least 1000 Hz or double the highest frequency present in the signal, to prevent aliasing (Stepp, 2012).

The effect of volitional swallowing manoeuvres may be very different when applied to healthy adults with optimised swallowing behaviours, compared to patients with dysphagia. The idea that there is a lower limit of muscle activity needed for swallowing suggests that patients with a sub-threshold level of activity would have insufficient motor unit recruitment for safe and functional swallowing. Further research investigating the effect of minimal effort swallowing on muscle activity in the dysphagic population will provide more insight into the clinical relevance of this manoeuvre.

In conclusion, the clinical application of this methodological study is that the sEMG biofeedback target for skill assessment and training should not be lower than 30% of maximum swallowing amplitude for healthy participants. Results show that healthy adults have the ability to modulate muscle activity, and this skill is retained regardless of age or bolus. Regular swallowing is more similar to minimal effort than maximal effort swallowing, reinforcing the idea of swallowing as a submaximal behaviour and challenging the traditional approach of maximal effort swallowing in dysphagia rehabilitation.

Chapter 7. Clinical classification of strength and movement-precision deficits in patients with dysphagia due to different aetiologies (Study 2)

7.1 Introduction

Swallowing is a complex behaviour that is regulated at multiple levels of neural control, including the central and peripheral nervous systems (Ertekin & Aydoğdu, 2003). At the peripheral level, the swallowing response involves the precise contraction and relaxation of multiple muscles that are innervated by cranial nerves. At central levels, the brainstem provides the basic, stereotyped sequence of swallowing events (Jean, 2001), and higher-level structures play a crucial role in the planning, coordination, and execution of motor response (Martin et al., 1999; Mosier & Bereznaya, 2001). By integrating sensory information from the bolus, cortical regions adapt the swallowing response to match environmental needs and allow for safe ingestion (Miller, 1999).

Neurological damage at any level can cause biomechanical impairments in the oral, pharyngeal, and/or oesophageal phases of swallowing (Daniels & Huckabee, 2014). The prevalence of strengthening treatments designed to target underlying muscle weakness in swallowing (Burkhead et al., 2007; Rogus-Pulia & Robbins, 2013) suggests an historical assumption that impaired biomechanical movement is caused mainly by decreased strength. However, evidence from the motor speech and limb literature indicates that cortical and peripheral lesions result in distinct patterns of pathophysiological features (Duffy, 2005; Kitago & Krakauer, 2010). Damage at the periphery generally causes reduced muscular force generation in the limbs, while cortical damage is characterised by impaired dexterity and weakness (Tomita & Usuda, 2013; Van Hedel et al., 2010; Wirth et al., 2008). Impaired dexterity in the limbs is the decreased coordination of skilled, voluntary movement to meet environmental demands (Ada et al., 1996). Given the precise coordination needed for safe swallowing, and the substantial role of the cortex in modulating the swallowing response, impairment in centrally-mediated movement precision has been proposed as a factor that underlies certain types of dysphagia (Daniels, 2000; Huckabee & Kelly, 2006; Huckabee et al., 2014; Huckabee & Macrae, 2014; Paik et al., 2008; Stepp et al., 2011).

While the effects of strength and movement precision impairments on limb functioning have been established (Canning et al., 2000), very few studies have investigated the relative contribution of strength and movement precision to swallowing. As previously discussed in Chapter 3, in this thesis document, the following definitions will be adhered to:

- *Impaired strength*, or *weakness* is decreased force generation that may be caused by several mechanisms, including direct changes to agonist motor units, secondary adaptive changes to muscle fibres, and indirect antagonist restriction of agonist activation.
- *Impaired movement precision* is the reduction in spatial and temporal accuracy of movement to meet environmental demands.

Recent research in the dysphagia literature has suggested that patients with dysphagia due to central damage may have decreased movement precision, with or without weakness (Paik et al., 2008). In one study, the extent and pattern of hyoid kinematic movement in patients with stroke and myopathy with dysphagia and healthy controls were examined using VFSS. Patients with myopathy had decreased hyoid range of motion and reduced epiglottic inversion, which were interpreted to indicate muscle weakness. On the other hand, stroke patients had adequate maximal hyoid displacement but a deviant spatial and temporal pattern of movement, suggesting incoordination. However, a limitation of VFSS is that the underlying cause of swallowing impairment can only be inferred, not directly measured, from visualisation of abnormal biomechanical movement (Clark, 2005). For example, decreased hyoid displacement seen on VFSS could be caused by reduced peripheral force generated by submental muscles, but it could also reflect centrally-mediated impairments in movement precision, impaired motor programming from cortical damage, or other neuromuscular deficits of motor control (Huckabee & Lamvik-Gozdzikowska, 2018). There is a need for the development of objective, measurable variables that can directly assess movement precision and strength.

Researchers have attempted to improve diagnostic specificity by investigating alternative assessment tools. sEMG is a non-invasive method of estimating muscle activity, and has been used in skill-based training and assessment protocols to target movement precision rather than maximal strength (Athukorala et al., 2014; Sella, 2012). The spatial and temporal aspects of muscle contraction can be displayed as a waveform on a screen in real-time, providing the patient with augmented biofeedback of their performance. Stepp, Britton, Chang, Merati, & Matsuoka (2011) investigated the ability of a video-game interface using neck sEMG biofeedback to assess movement precision. A stroke patient with dysphagia and healthy controls controlled the timing and submaximal force of any available neck movement (not just swallowing) to place the response cursor inside an on-screen target. The sEMG biofeedback was calibrated to each participant's maximum muscle activity measured during effortful swallowing. The patient with dysphagia acquired substantially fewer targets compared to the

healthy controls, suggesting that “hit rate” (percentage of total targets that are acquired) could be a feasible measure of voluntary control and precision. However, limited generalisation to swallowing behaviour in the patient population can be made from the results of a single patient, who controlled muscle activation biofeedback using behaviour not specific to swallowing. In addition, valuable information regarding temporal and amplitude precision may not be captured with a measure of hit rate. Swallowing precision may be further quantified using the temporal and amplitude errors between the target and response for submaximal attempts. Finally, even though calibration allowed individuals to participate in the task within the limits of their available muscle activity, the study did not directly measure their strength levels, leading to the possibility that weakness may have confounded the results.

Most research investigating objective measures of weakness in swallowing has focused on lingual musculature (Butler et al., 2011; Clark et al., 2003; Yoshida et al., 2006). However, little is known regarding the relationship between submental muscle strength and swallowing function. The submental muscles are important for hyolaryngeal excursion during swallowing and also for opening the jaw. Since decreased hyoid displacement is associated with greater risk of airway invasion and pharyngeal residuals (Steele et al., 2011), decreased submental strength and contraction may contribute towards dysphagia. Submental strength can be indirectly estimated using maximum sEMG activity during effortful swallowing. A more direct measure of strength can be achieved using muscle dynamometry, which has been used in the physical therapy domain for valid and reliable measures of maximum force generated by a muscle or muscle group (Bohannon, 1986, 2007). A jaw-opening force test, using a dynamometer placed under the chin and secured using a head strap, has been developed to assess submental muscle strength (Tohara et al., 2011). Jaw-opening force was significantly different between healthy older and younger adults (Iida et al., 2013), and has been found to be related to the size of submental muscles (Kajisa et al., 2018). Measurement of jaw-opening force was found to have high sensitivity and specificity for predicting pharyngeal residuals but not aspiration (Hara et al., 2014), suggesting that patients with dysphagia may have other impairments besides only weakness. Therefore, a clinical assessment of the underlying mechanisms of dysphagia might combine measurements of both strength and movement precision in the submental muscles.

The objective of the study was to clinically classify healthy controls and patients with dysphagia due to stroke and myopathy into subgroups based on a novel, objective assessment of swallowing-related strength and movement precision. The performance of submental

muscles during both swallowing and jaw-opening was quantified with four assessment tasks using dynamometry and sEMG. An unsupervised clustering method was used to uncover any performance patterns or participant subgroups. The ability of the assessment to discriminate between patterns of strength and movement precision was also evaluated by comparing test performance between patients with dysphagia after stroke, patients with dysphagia due to myopathy, and healthy controls. It was hypothesised that stroke patients would have decreased performance on measures of both strength and movement precision, while patients with myopathy would have deficits in measures of strength only. This exploratory study takes the first step in differentiating dysphagia caused by weakness from other pathophysiology, thereby setting the foundation for future research into improving diagnostic specificity.

7.2 Methodology

7.2.1 *Participants*

There are no established guidelines for calculating sample size for cluster analyses (Dolnicar, 2002). However, after consultation with a statistician, it was estimated that a sample size of approximately 20 participants would be appropriate for each cluster. Since healthy controls and patients with myopathy were expected to be assigned into their own clusters, and stroke patients were expected to be assigned to several clusters, a larger number of stroke patients was needed. Therefore, recruitment of 20 healthy participants, 20 patients with myopathy, and 60 stroke patients was targeted. Recruitment of healthy participants was later increased to 40 to ensure a normative sample with adequate age and sex representation.

Healthy participants aged 50 or older were recruited using a written advertisement sent to individuals on the University of Canterbury Rose Centre for Stroke Recovery and Research participant database. Exclusion criteria included a presence or history of swallowing disorders, neuromuscular dysfunction, or temporomandibular joint disorders or surgery. Patients aged 50 or older who had dysphagia due to stroke or myopathy (inclusion body myositis, myotonic dystrophy, or oculopharyngeal muscular dystrophy) were also recruited. This was accomplished by sending a written advertisement to the University of Canterbury Rose Centre participant database, the Muscular Dystrophy Association of New Zealand Registry, the Stroke Foundation of New Zealand, rest homes, and the speech-language therapy departments at five District Health Board (DHB) hospitals in New Zealand (Canterbury, Waitemata, Capital and Coast, Hutt Valley, and Wairarapa). Potential participants contacted the researcher directly, or

were referred by a healthcare professional from the recruitment sites. An appointment was made with potential participants either at the University of Canterbury Rose Centre for Stroke Recovery and Research, or at the participant's residence (home, hospital, or rest home) if requested.

At the beginning of the appointment, a questionnaire regarding medical history was provided to ensure participants met inclusion/exclusion criteria. Presence of clinically-detectable dysphagia was confirmed in the stroke and myopathy patients using the EAT-10, a patient-centred questionnaire that measures self-reported symptoms of dysphagia (Belafsky et al., 2008), and a clinical swallowing examination. The clinical swallowing examination included examination of the oral mechanism and cranial nerves, as well as an assessment of oral intake using the Timed Water Swallow Test (TWST; Hughes & Wiles, 1996) and Test of Mastication and Swallowing Solids (TOMASS; Huckabee et al., 2018). For the patient's safety, the oral intake assessment was tailored to each patient according to their level of swallowing impairment, and their ingestion was monitored carefully by the researcher. Written informed consent was obtained prior to data collection, and the study was approved by the appropriate regional Human Ethics Committee (see Appendices).

7.2.2 Instrumentation

7.2.2.1 sEMG and BiSSkiT software

Submental sEMG signals were obtained using a circular, self-adhesive, disposable electrode patch (EMG Triode™ Electrode, Thought Technology Ltd., Canada). Three electrodes (two recording electrodes and one ground) were embedded in the patch, equidistant to each other in a triangular pattern. Inter-electrode distance was 2 cm between the centre of each electrode and 1 cm between the lateral edges. The skin surface under the chin was prepared prior to electrode placement in order to improve skin-electrode contact, using repetitive placement and removal of adhesive tape on the skin for light abrasion, followed by skin cleansing with an alcohol wipe (Merletti & Parker, 2004; Stepp, 2012). The electrode patch was attached to the skin surface underneath the chin to measure the activity of the collective submental muscle group (Figure 7.1). The two recording electrodes were placed at midline, overlying the submental muscles, with the first electrode approximately 2 cm posterior to the anterior, inferior midline of the mandible and the other electrode 1 cm posterior to the first one. The ground electrode was oriented lateral to midline. Since the surface electrodes were placed over the floor-of-mouth

muscles, electrical activity was measured from the collective anterior bellies of the digastric, mylohyoid, and geniohyoid muscles (Palmer et al., 1999).

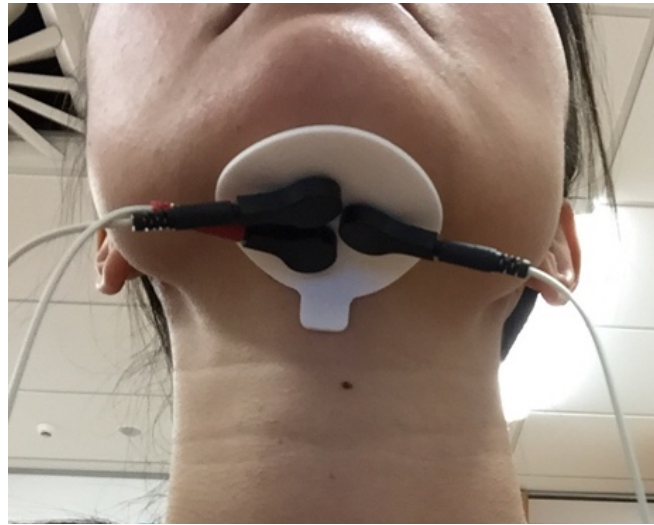


Figure 7.1. Placement of the adhesive electrode patch on the surface of the skin under the chin for submental sEMG.

sEMG signals were recorded by a portable sEMG device (NeuroTrac® Simplex, Verity Ltd., UK). Data were sent via a fibreoptic cable to a USB serial port which was plugged into a laptop operating the BiSSkiT software. The signal was used to plot a real-time waveform on the laptop screen, with time in seconds on the x-axis and amplitude in μV on the y-axis. A significant change in the activity of the submental muscle group (e.g., when swallowing or mouth-opening) was typically depicted on the screen as a peak in the waveform. The laptop was placed on a table in view of the participant, with the top of the laptop screen adjusted to the participant's eye level. Participants were instructed to keep their head as still as possible during measurement of sEMG activity. Data from BiSSkiT software was saved to a .csv file and analysed offline on a personal computer.

7.2.2.2 Dynamometry

Jaw-opening force was measured using a compact dynamometer (Commander PowerTrack, JTech, USA) placed under the chin (Hara et al., 2014). The dynamometer was secured to the chin and head using a custom-made device consisting of polyester webbing straps, molded chin cup, and a baseball cap (Figure 7.2). A molded chin cup made of dental putty was attached to the dynamometer sensor plate. The chin cup allowed the dynamometer to be securely held in place under the participant's chin. The cap band was tightened around the participant's head

using the hook and loop back closure, to prevent extraneous movement. The vertical straps were adjusted on either side of the head using two plastic cam buckles so that the dynamometer was held as tightly as possible under the chin, to prevent the jaw from opening. Data were sent from the dynamometer to a hand-held controller, which displayed the magnitude of force in real-time.



Figure 7.2. Jaw-opening force test with dynamometer secured under the chin, and hand-held controller displaying force in Newtons.

7.2.3 Experimental procedure

Written informed consent was obtained. Demographic information was collected from patient report and/or medical records. The patient's diet level was categorised as nil by mouth (NBM; receiving the majority of nutrition via non-oral means), modified diet (thickened liquids and/or minced/moist/pureed solids), or regular diet (soft, bite-sized, or regular solids and thin liquids). Regional Health and Disability ethical approvals were received for this prospective experimental study.

All participants completed four assessment tasks measuring swallowing strength, swallowing movement precision, jaw-opening strength, and jaw-opening movement precision. The four

tasks were counter-balanced to prevent order effects. All swallows were performed with saliva only (without food or liquid). Participants were allowed to have sips of water or other drinks between tasks if they requested it and could ingest it safely.

7.2.3.1 Swallowing strength assessment

The participant was trained to perform effortful swallowing, given the verbal directions, “Swallow hard with all the muscles in your mouth and throat”. During training, they were provided with submental sEMG biofeedback on the computer screen, and encouraged to make the waveform peak go as high as possible with effortful swallowing. When the participant understood and demonstrated the task appropriately, as evidenced by peak amplitude of effortful swallowing substantially higher than peak amplitude of regular swallowing, data collection began. The participant was instructed to perform five effortful swallows, and then five regular effort swallows, at a rate of approximately one every 30 seconds. No visual feedback was provided to the participant during data collection.

7.2.3.2 Swallowing movement-precision assessment

The average peak amplitude of the five effortful swallows from the swallowing strength assessment was detected by the software to represent the calibration value. The y-axis was adjusted so that the maximum amplitude equalled the calibration value. A square target appeared in the centre of the waveform display (see Figure 7.3) and did not change size or shape throughout assessment. When used for movement precision *training*, the biofeedback software varied target size and shape at each trial, as the objective was to increase task challenge and optimise movement precision learning over time. However, for movement precision *assessment*, the size and shape of the target was kept constant to allow for comparison of task performance between participants. Since the height of the target box was 30% of the y-axis height and the target was placed in the centre of the screen, the lower and upper limit of the box was 35% and 65% of the y-axis respectively. This lower limit was chosen because 30% of one’s maximum muscle activity is the minimum amount of contraction needed to initiate swallowing (from Study 1). Lowering the threshold any further would make it unlikely for participants to acquire the target, thus confounding results. The upper limit was chosen because a higher threshold would result in the task more closely resembling an effortful strength-based assessment of swallowing, instead of a submaximal movement precision assessment. The participant was instructed to watch the screen and “swallow so that the peak of your waveform

falls in the centre of the square.” Participants completed 10 trials. Inter-trial interval was approximately 30 seconds.

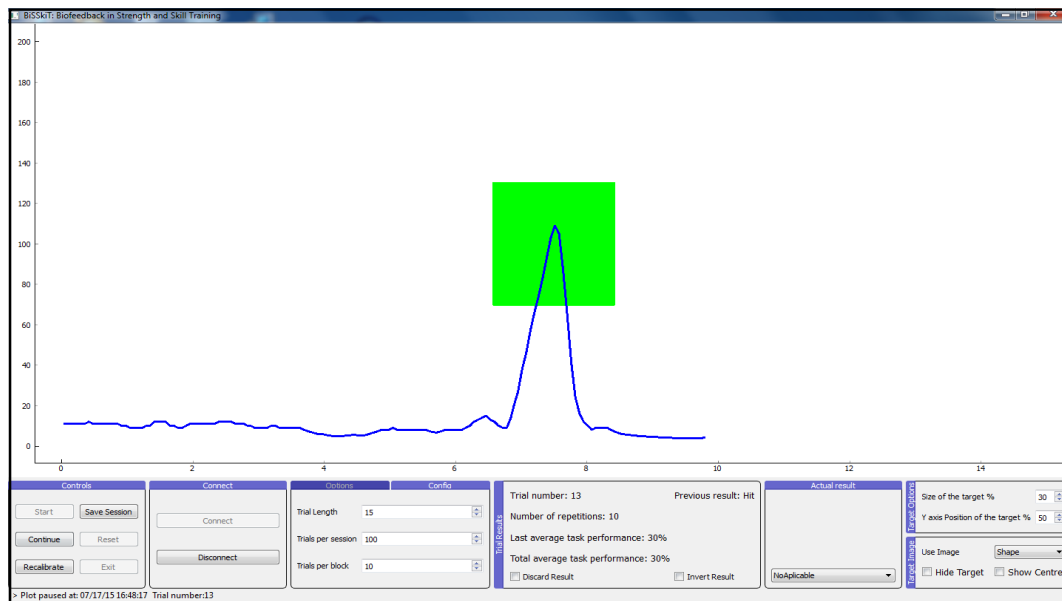


Figure 7.3. Screenshot of BiSSkiT software during movement precision assessment.

7.2.3.3 Jaw-opening strength assessment

The dynamometer was calibrated to zero, and then secured to the participant's head and chin. The participant was given the instructions: "This test will measure your maximum strength as you open your jaw. I would like you to gradually increase your jaw-opening force over 1 second until you reach your maximum force. Hold for 2 seconds, and then relax." The controller displayed the maximum force generated at each trial. The examiner wrote down the value and then cleared it from device memory in preparation for the next trial. Participants were not provided with biofeedback of their jaw-opening force. Participants completed five trials, with a break of at least 1 minute between trials. The straps were loosened between trials, and re-tightened before resuming data collection.

7.2.3.4 Jaw-opening movement precision assessment

Using submental sEMG, the mean peak amplitude from five trials of maximum jaw-opening was calculated as the calibration value. The y-axis was then adjusted so that the maximum amplitude equalled the calibration value. As in the swallowing movement precision assessment, a square target appeared in the centre of the waveform display. The height and width of the target was equal to 30% of the screen height. Target size and position remained

constant over 10 trials. The participant was instructed to “open your jaw so that the peak of the waveform falls in the centre of the square.” Inter-trial interval was approximately 30 seconds.

7.2.4 Outcome measures

The raw data were analysed to produce the following eight strength and movement precision outcome measures for each participant. A “hit” during movement precision tasks was defined as the waveform peak falling inside the square target box. Peak-to-target error was defined as the distance between the centre of the target and the peak of the waveform (Figure 7.4). A smaller error represented increased accuracy/precision.

Swallowing strength (sEMG)

1. Normalised effortful swallowing amplitude (ES): Mean sEMG peak amplitude of effortful swallows, divided by mean sEMG peak amplitude of regular effort swallows, expressed as a ratio

Jaw-opening strength (Dynamometer)

2. Jaw-opening force (JF): Mean jaw-opening force from five trials, in Newtons

Swallowing movement-precision (sEMG with biofeedback)

3. Swallowing hit rate (SHR): Frequency of hits during swallowing movement precision task, expressed as a percentage
4. Swallowing relative temporal error (STE): Mean peak-to-target temporal error in seconds divided by width of the screen (30 seconds), expressed as a percentage
5. Swallowing relative amplitude error (SAE): Mean peak-to-target amplitude error in μV divided by height of the screen (calibration value in μV), expressed as a percentage

Jaw-opening movement-precision (sEMG with biofeedback)

6. Jaw-opening hit rate (JHR): Frequency of hits during jaw-opening movement precision task, expressed as a percentage
7. Jaw-opening relative temporal error (JTE): Mean peak-to-target temporal error in seconds divided by width of the screen (30 seconds), expressed as a percentage

8. Jaw-opening relative amplitude error (JAE): Mean peak-to-target amplitude error in μV divided by height of the screen (calibration value in μV), expressed as a percentage

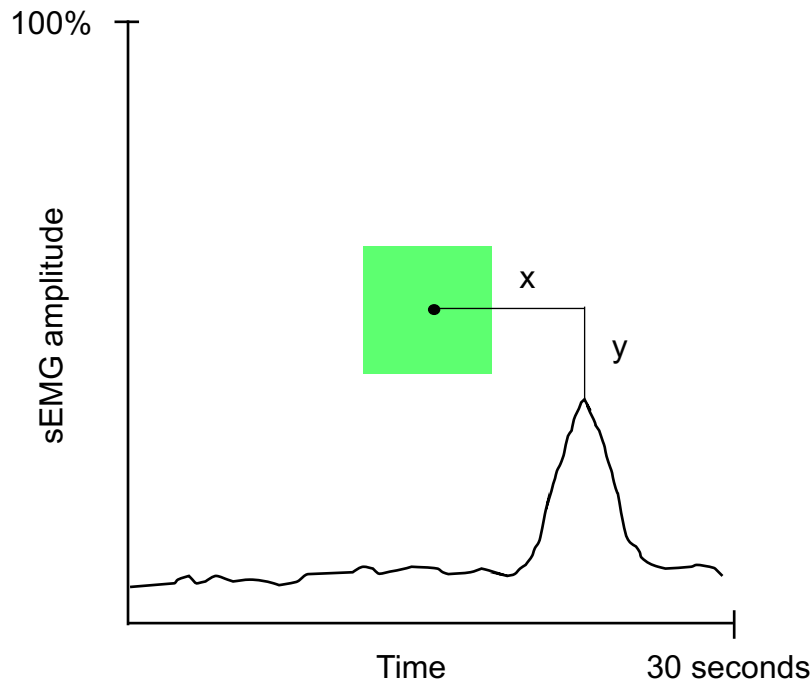


Figure 7.4. Computation of peak-to-target error as a measure of accuracy for movement precision tasks. Relative temporal error is the time x between the centre of the target and response peak, divided by 30 seconds. Relative amplitude error is the difference in amplitude between the centre of the target and response peak (y), divided by the calibration value.

7.2.5 Statistical analysis

Variance inflation factors (VIFs) were calculated to test for multicollinearity between the strength and movement precision variables. Variables with a VIF threshold value above 10 were considered to have over-inflated variances (Kutner, Nachtsheim, Neter, & Li, 2005) and not included in further analyses.

7.2.5.1 Classification of test performance based on diagnostic groups

To determine differences in performance on the strength and movement precision assessment between the three diagnostic groups, a multivariate analysis of variance (MANOVA) was run on the continuous variables (normalised effortful swallowing amplitude, jaw force, swallowing temporal error, swallowing amplitude error, jaw temporal error, and jaw amplitude error). If

significant, univariate one-way analyses of variance (ANOVAs) were conducted on each of the continuous variables. For the non-continuous variables (swallowing and jaw hit rate) generalised linear mixed effect models for binomial distributions were completed, with diagnostic group as the fixed effect and participant as random effect. For each variable demonstrating a significant effect of group, follow-up Tukey post-hoc tests were performed to determine pairwise differences.

7.2.5.2 Classification of test performance with unknown groups

To determine if there were clusters of participants based on their strength and movement precision assessment performance, a hierarchical cluster analysis using Ward's method and Euclidean distance was completed. Cluster analysis partitions participants into clusters by maximizing similarities within groups and differences between groups (Tan, Steinbach, & Kumar, 2005). Using this approach, each participant is initially considered to be its own cluster. Clusters are progressively combined based on their similarity, and this step is repeated until all participants are members of one cluster. Cluster validation indices can then be used to determine the optimal number of clusters for a given solution, by evaluating intracluster compactness and intercluster separation. Given the exploratory nature of cluster analysis, there is no single accepted index used to determine the optimal number of clusters (Kaufman & Rousseeuw, 1990). It was decided that a minimum of three clusters was needed because there were three diagnostic groups. Twenty-six different indices were then calculated, and the optimal number of clusters was decided by majority rule (Charrad, Ghazzali, Boiteau, & Niknafs, 2014). Comparison of the association between cluster and diagnostic group membership was analysed using Fisher's exact test.

Relative performance differences between the clusters were investigated using a MANOVA and univariate ANOVAs on the continuous variables, and a generalised linear mixed effect model for binomial distributions on the binomial variables (cluster as fixed effect and participant as random effect). Post-hoc Tukey HSD tests were conducted to examine to determine pairwise differences. It is important to note that the analyses of variance comparing clusters based on strength and movement precision variables were used to describe relative effect sizes and not calculate statistical significance, as the clusters were formed based on maximising Euclidean distance between the same variables.

To evaluate the internal validity of the cluster solution, the cohesion and separation of the clusters were visualised on a scatterplot using the first two principal components, with

participants presented as individual points on the plot. A silhouette analysis was conducted to provide an objective measure of how well each participant was clustered (Kaufman & Rousseeuw, 1990). The silhouette width is a measure of distance between clusters, and ranges from -1 (poorly clustered) to 1 (well-clustered). To evaluate the external validity of the cluster solution, the performance on the TWST and TOMASS was compared between clusters using a MANOVA and univariate ANOVAs on the continuous variables, and generalised linear mixed effect models for binomial distributions with cluster as fixed effect and participant as random effect for the binary variables.

To evaluate if any of the eight strength and movement precision variables could be reduced, the relationship between the variables were investigated using Spearman's rank correlation coefficients. Significance values were calculated using permutation tests to control for multiple comparisons. The classification and regression tree (CART) technique was used to identify which variables best predict a participant's strength and movement precision performance, and the threshold cut-off values used for decision-making. The classification tree was modelled with assigned cluster as the target (dependent) variable and the eight strength and movement precision variables as the predictor (independent) variables, using the Gini splitting criterion. The trees were then pruned to avoid overfitting of the data, using the complexity parameter that minimised cross-validated error.

7.3 Results

A total of 133 participants were recruited for the study. Eleven individuals (four recruited for the healthy group, five in the stroke group, and two in the myopathy group) were excluded because they did not meet inclusion/exclusion criteria. Specifically, excluded individuals had a history of neuromuscular dysfunction other than stroke or myopathy (n=5), were under the age of 50 (n=2), and were stroke patients without clinically-detectable dysphagia (n=4). In addition, eight stroke patients initiated but were unable to complete the study, due to fatigue (n=2), severity of dysphagia and inability to initiate volitional swallowing (n=4), and severity of cognitive impairments and inability to follow directions (n=2). One hundred and fourteen participants were included in the final analyses (40 healthy, 55 stroke, and 19 myopathy participants).

None of the strength and movement precision variables exceeded the suggested VIF multicollinearity threshold of 10 (Kutner et al., 2005), therefore all the variables were kept in the analyses (Table 7.1).

Table 7.1

Variance Inflation Factors for Strength and Movement Precision Variables

Variable	Variance inflation factor
Effortful swallowing	1.2
Jaw force	1.1
Swallowing hit rate	4.0
Swallowing temporal error	3.3
Swallowing amplitude error	2.6
Jaw hit rate	4.6
Jaw temporal error	2.6
Jaw amplitude error	3.5

7.3.1 Comparison of diagnostic groups

While the three diagnostic groups did not differ on demographic variables of sex or ethnicity (Table 7.2), stroke patients were older than both the healthy participants ($p < .001$) and myopathic patients ($p < .001$). Dysphagic characteristics of the stroke and myopathy patients are displayed in Table 7.3. Significant differences were noted in terms of the acuity of dysphagia onset (as would be expected by nature of the disorder), diet level, and EAT-10 scores.

Table 7.2

Participant Demographics by Diagnostic Group

	Healthy (n = 40)	Stroke (n = 55)	Myopathy (n = 19)	<i>p</i> value
Age (years):				<.001
Mean \pm SD	69.0 \pm 9.91	78.4 \pm 9.28	64.6 \pm 8.75	
Range	51 – 88	55 – 94	52 – 80	
Female	20 (50%)	18 (33%)	11 (58%)	.09

Ethnicity:				.40
Caucasian	37 (93%)	51 (93%)	19 (100%)	
New Zealand Maori	0 (0%)	2 (3%)	0 (0%)	
Pacific Islander	0 (0%)	1 (2%)	0 (0%)	
Other	3 (7%)	1 (2%)	0 (0%)	
Stroke lesion location:	N/A		N/A	N/A
Supratentorial – Right		19 (35%)		
Supratentorial – Left		17 (31%)		
Infratentorial		9 (16%)		
Unknown		10 (18%)		
Myopathy type:	N/A	N/A		N/A
Myotonic dystrophy			12 (63%)	
Inclusion body myositis			6 (32%)	
Oculopharyngeal muscular dystrophy			1 (5%)	

Note. SD = standard deviation.

Table 7.3

Dysphagia Characteristics of Stroke and Myopathy Groups

	Stroke (n = 55)	Myopathy (n = 19)	<i>p</i> value
Dysphagia duration:			<.001
Acute (≤3 months)	25 (45%)	0 (0%)	
Chronic (>3 months)	30 (55%)	19 (100%)	
Oral intake status:			.03
Non-oral	4 (7%)	1 (5%)	
Modified oral	23 (42%)	2 (11%)	

Full oral	28 (51%)	15 (79%)	
EAT-10 score (Mean \pm SD)	12.1 \pm 8.42	18.3 \pm 8.86	.01
TWST score (Mean \pm SD)			
Volume/swallow	13.82 \pm 7.56	18.95 \pm 9.17	.05
Time/swallow	3.83 \pm 2.68	3.52 \pm 3.00	.72
Volume/time	5.54 \pm 5.05	7.79 \pm 5.37	.16
TOMASS score (Mean \pm SD)			
Bites	3.68 \pm 1.94	4.19 \pm 1.64	.35
Masticatory cycles	101.19 \pm 45.86	93.75 \pm 35.55	.54
Swallows	2.77 \pm 1.73	4.69 \pm 3.36	.05
Time	97.47 \pm 44.34	101.05 \pm 33.16	.76

Note. SD = standard deviation, EAT-10 = 10-Item Eating Assessment Tool, TWST = Timed Water Swallowing Test, TOMASS = Test of Masticating and Swallowing Solids.

Table 7.4 summarises the strength and movement precision assessment results for the three groups. There was high inter-individual variability within groups, particularly for stroke patients on the movement precision variables. Multivariate analysis demonstrated that the stroke, myopathic and healthy groups performed differently from each other on the six continuous strength and movement precision variables [Pillai's trace = .68, $F(2, 111) = 9.26$, $p < .001$]. Univariate analyses conducted on all eight variables showed an effect of diagnostic group for normalised effortful swallowing amplitude [$F(2, 111) = 12.6$, $p < .001$], jaw force [$F(2, 111) = 22.47$, $p < .001$], swallowing hit rate [$\chi^2(2) = 25.84$, $p < .001$], swallowing temporal error [$F(2, 111) = 30.67$, $p < .001$], jaw opening hit rate [$\chi^2(2) = 24.68$, $p < .001$], jaw temporal error [$F(2, 111) = 22.81$, $p < .001$], and jaw amplitude error [$F(2, 111) = 7.98$, $p < .001$]. There was no effect of diagnostic group for swallowing amplitude error [$F(2, 111) = 20.76$, $p = .65$].

Table 7.4

Mean Outcomes \pm SD on Strength and Movement Precision Assessment by Diagnostic Group

Variable	Healthy	Stroke	Myopathy
----------	---------	--------	----------

ES (ratio)	3.0 ± 1.7	1.7 ± 0.9	1.9 ± 0.7
JF (N)	93.8 ± 26.4	70.6 ± 22.4	52.1 ± 20.8
SHR (%)	53.5 ± 21.8	31.5 ± 22.2	54.6 ± 25.3
STE (%)	2.1 ± 1.2	9.0 ± 6.1	3.3 ± 2.6
SAE (%)	16.4 ± 6.1	17.3 ± 8.0	15.7 ± 5.8
JHR (%)	69.6 ± 15.0	47.8 ± 24.5	57.4 ± 17.9
JTE (%)	1.4 ± 0.6	3.9 ± 2.5	2.0 ± 1.0
JAE (%)	12.2 ± 3.3	17.0 ± 7.4	15.0 ± 4.1

Note. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error, N = Newtons.

Further post-hoc analyses were completed to investigate differences between groups on each strength and movement precision variable (Table 7.5). Participants in both the stroke and myopathy groups had reduced amplitude during effortful swallowing and jaw force compared to healthy controls. Stroke patients had impaired performance on all movement precision tasks compared to healthy controls, except for swallowing amplitude error. Stroke patients also had impaired movement precision performance compared to myopathy patients on three tasks, namely decreased swallowing hit rate and increased swallowing and jaw temporal errors. There was no difference on movement precision task performance between healthy and myopathy groups.

Table 7.5

Results of Post-Hoc Analysis Comparing Mean Differences (95% CI) Between Groups

	Healthy – stroke	Healthy – myopathy	Myopathy – stroke
ES (ratio)	1.2** (0.6 – 1.8)	1.0** (0.3 – 1.8)	0.2 (-0.6 – 0.9)
JF (N)	23.2** (11.6 – 34.9)	41.7** (26.1 – 57.4)	-18.5** (-33.4 – -3.5)
SHR (%)	1.1** (0.6 – 1.5)	-0.1 (-0.6 – 0.5)	1.1** (0.5 – 1.7)
STE (%)	-6.9** (-9.1 – -4.7)	-1.2 (-4.1 – 1.8)	-5.7** (-8.5 – -2.9)

SAE (%)	-0.9 (-4.4 – 2.6)	0.7 (-3.9 – 5.4)	-1.6 (-6.1 – 2.8)
JHR (%)	1** (0.6 – 1.4)	0.6 (0.1 – 1.1)	0.4 (-0.1 – 0.9)
JTE (%)	-2.4** (-3.3 – -1.6)	-0.6 (-1.8 – 0.6)	-1.8** (-3.0 – -0.7)
JAE (%)	-4.8** (-7.6 – -1.9)	-2.7 (-6.5 – 1.1)	-2.0 (-5.7 – 1.6)

Note. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

7.3.2 Hierarchical cluster analysis

Results of the hierarchical cluster analysis are displayed in a dendrogram (Figure 7.5). According to the 26 different cluster validation indices that were calculated, five indices proposed three as the best number of clusters, nine proposed four clusters, and four proposed seven clusters (Figure 7.6). According to majority rule, the most frequently proposed number of clusters was four.

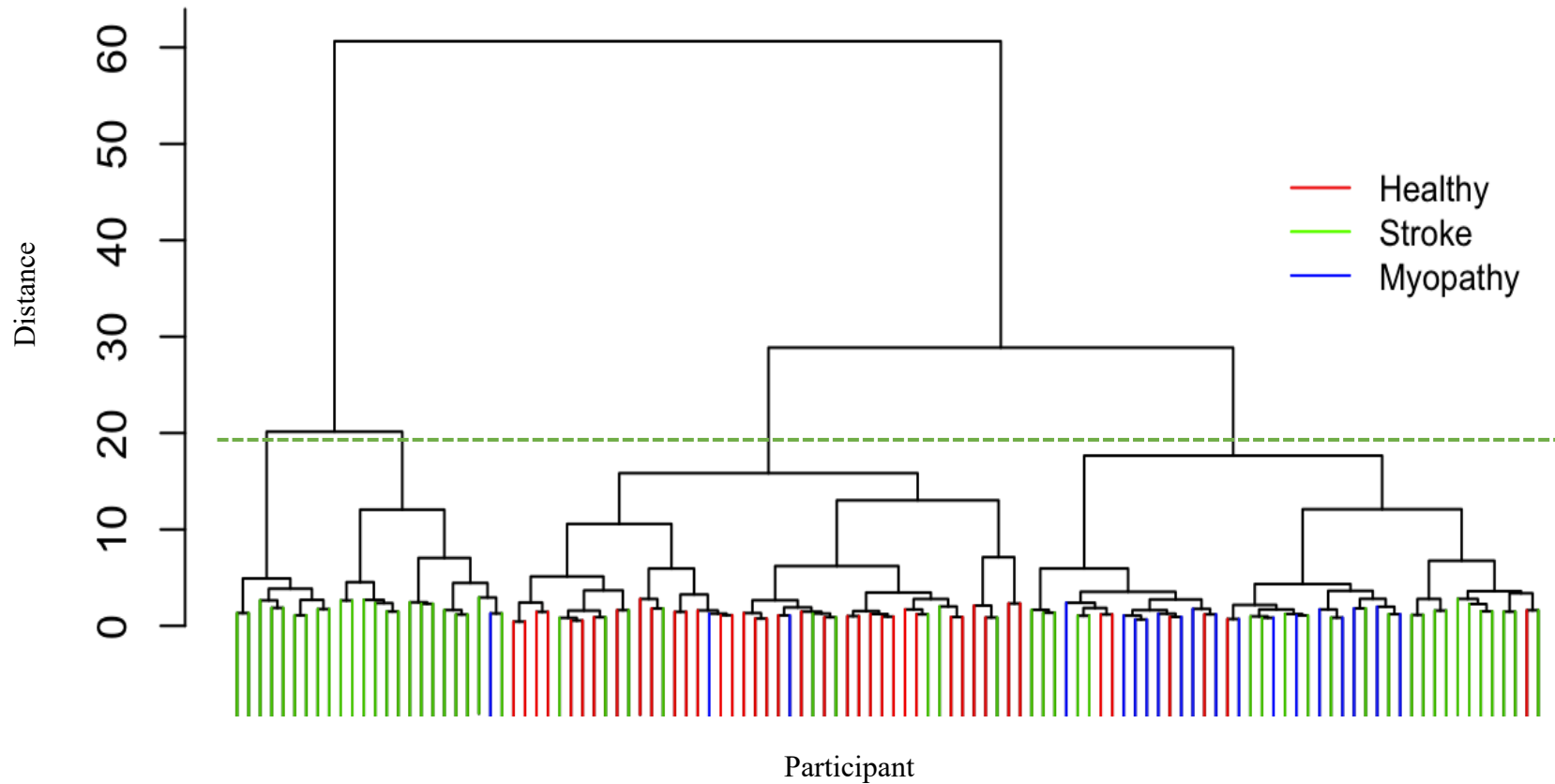


Figure 7.5. Clustering of healthy, stroke, and myopathy participants (coded by colour) using hierarchical cluster analysis. Each “leaf” at the bottom represents a participant. The vertical “branches” reflect the distance/dissimilarity at which clusters of participants were merged. Cutting of the dendrogram branches at the dashed line results in four clusters. The optimal number of four clusters was calculated using 26 different methods to determine the most frequently proposed number of clusters.

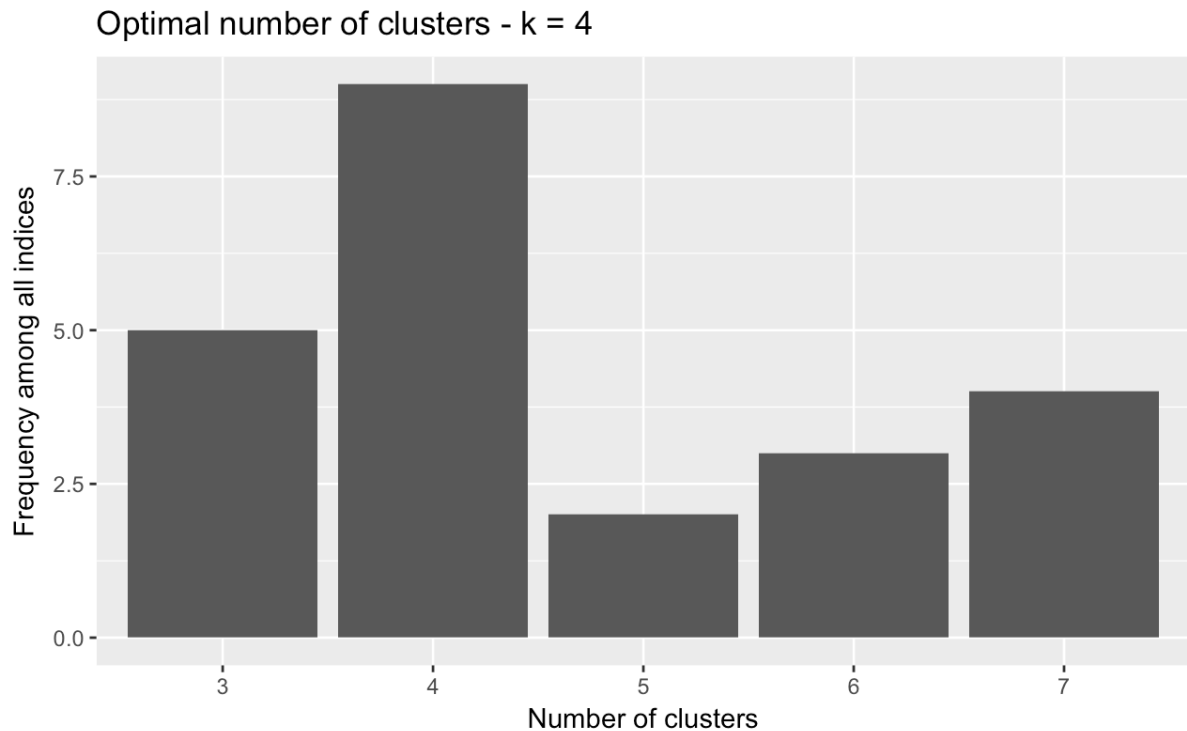


Figure 7.6. Frequency at which different numbers of clusters were recommended by 26 cluster validation indices.

Characteristics of the four clusters are summarized in Table 7.6. Membership in diagnostic group and cluster were significantly associated according to the Fisher exact test, $p < .001$. While most of the healthy (85%) and myopathic (84%) participants were assigned to unique clusters, the stroke patients were spread across the four clusters. There was an overall effect of cluster on the strength and movement precision outcomes [Pillai's trace = 1.58, $F(3, 110) = 19.97$, $p < .001$]. Univariate analyses conducted on each strength and movement precision variable showed an effect of cluster for normalised effortful swallowing amplitude [$F(3, 110) = 11.21$, $p < .001$], jaw force [$F(3, 110) = 23.93$, $p < .001$], swallowing hit rate [$\chi^2(3) = 62.57$, $p < .001$], swallowing temporal error [$F(3, 110) = 43.46$, $p < .001$], swallowing amplitude error [$F(3, 110) = 20.63$, $p < .001$], jaw opening hit rate [$\chi^2(3) = 110.26$, $p < .001$], jaw temporal error [$F(3, 110) = 49.07$, $p < .001$], and jaw amplitude error [$F(3, 110) = 43.63$, $p < .001$].

Table 7.6

Diagnostic Group Membership and Strength/Movement Precision Assessment Outcomes, Stratified by Cluster

		Cluster			
		1	2	3	4
Diagnosis	Healthy	34 (85%)	0 (0%)	6 (15%)	0 (0%)
	Stroke	9 (16%)	14 (25%)	23 (42%)	9 (16%)
	Myopathy	2 (11%)	1 (5%)	16 (84%)	0 (0%)
Strength and movement precision	ES (ratio)	3.0 ± 1.7	1.4 ± 0.6	1.9 ± 0.7	1.5 ± 0.7
	JF (N)	97.1 ± 25.6	64.0 ± 20.0	60.3 ± 20.0	65.0 ± 11.1
	SHR (%)	49.4 ± 19.9	20.7 ± 15.3	51.6 ± 23.4	5.6 ± 10.1
	STE (%)	2.4 ± 1.5	14.2 ± 5.7	4.9 ± 4.33	11.2 ± 5.0
	SAE (%)	17.1 ± 6.0	16.7 ± 5.5	13.7 ± 5.4	30.1 ± 6.0
	JHR (%)	71.9 ± 14.6	15.3 ± 13.0	55.9 ± 14.0	57.8 ± 13.0
	JTE (%)	1.4 ± 0.5	6.1 ± 3.2	2.4 ± 1.0	4.8 ± 1.7
	JAE (%)	11.9 ± 3.3	25.9 ± 7.3	14.9 ± 3.8	12.6 ± 3.0

Note. Diagnostic group data presented as counts (and percentage of the diagnostic group), and assessment data presented as means ± SD. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

Post-hoc comparisons between clusters, and comparisons to the average scores across all participants, showed that the four clusters demonstrated characteristic patterns of performance (Table 7.7 and Figure 7.7).

- Cluster 1. This cluster was mainly made up of healthy participants. It had the best strength outcomes compared to other clusters (highest effortful swallowing amplitude and jaw force values). Participants in this cluster also demonstrated good movement precision outcomes relative to the other participants.

- Cluster 2. Most participants in cluster 2 were stroke patients. Cluster 2 was characterised by decreased strength and movement precision, with all variables (except for swallowing amplitude error) below the average. Cluster 2 had the most impaired jaw movement precision scores of all the clusters, with the lowest jaw hit rate and highest jaw peak-to-target error.
- Cluster 3. This cluster comprised stroke, myopathy, and healthy participants. It had below-average effortful swallowing amplitude and jaw force that was lower than cluster 1, but comparable to clusters 2 and 4. Swallowing amplitude error and swallowing hit rate values were similar to cluster 1. Jaw temporal and amplitude error and swallowing temporal error were mildly impaired compared to cluster 1, but still at or above the mean, and better than clusters 2 and 4.
- Cluster 4. All participants in cluster 4 were stroke patients. This cluster had the most impaired scores on swallowing hit rate and amplitude error. However, jaw hit rate and temporal error scored at or above average.

Table 7.7

Comparison of Differences Between Clusters for Strength and Movement Precision Variables

Variable	Cluster pairing					
	2 – 1	3 – 1	4 – 1	3 – 2	4 – 2	4 – 3
ES (ratio)	-1.6**	-1.1**	-1.5**	0.4	0.1	-0.4
JF (N)	-33.1**	-36.9**	-32.2**	-3.8	0.9	4.7
SHR (%)	-28.7**	2.2	-43.8**	31.0**	-15.1*	-46.1**
STE (%)	11.8**	2.5*	8.8**	-9.3**	-3.0	6.3**
SAE (%)	-0.4	-3.4*	13.0**	-3.0	13.3**	16.3**
JHR (%)	-56.6**	-15.9**	-14.1*	40.6**	42.5**	1.8
JTE (%)	4.7**	1.0**	3.4**	-3.7**	-1.3	2.4**
JAE (%)	14.3**	3.0**	0.7	-11.1**	-13.3**	-2.3

Note. p-values for descriptive purposes only. *p < .05. **p < .01. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

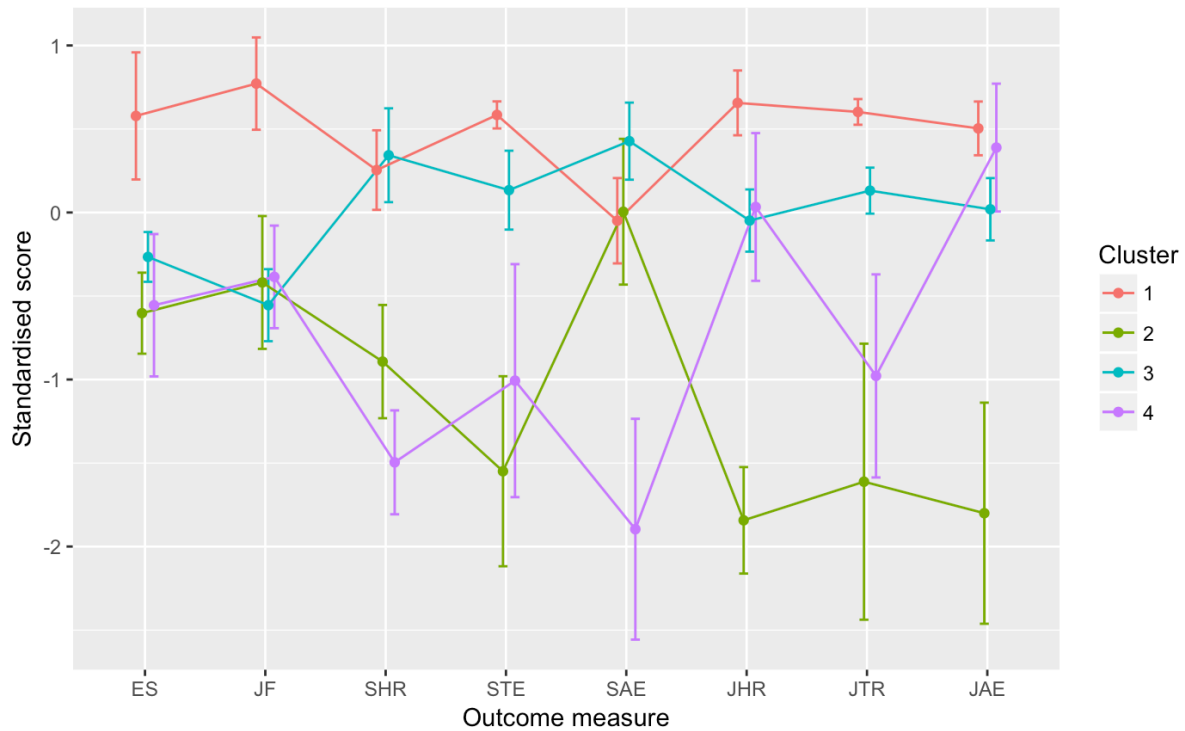


Figure 7.7. Standardised mean scores and 95% confidence intervals for strength and movement precision variables. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error. Scores for SAE, STE, JAE, and JTE have been reversed for ease of interpretation, so that a score below zero reflects greater impairment.

7.3.2.1 Evaluation of cluster solution

Visual inspection of the principal components scatterplot (Figure 7.8) shows separation between all the clusters, except for some overlap between clusters 1 and 3. Average silhouette width was .14 for cluster 1, .23 for cluster 2, .09 for cluster 3, and .36 for cluster 4, with an overall mean silhouette width across all clusters of .17.

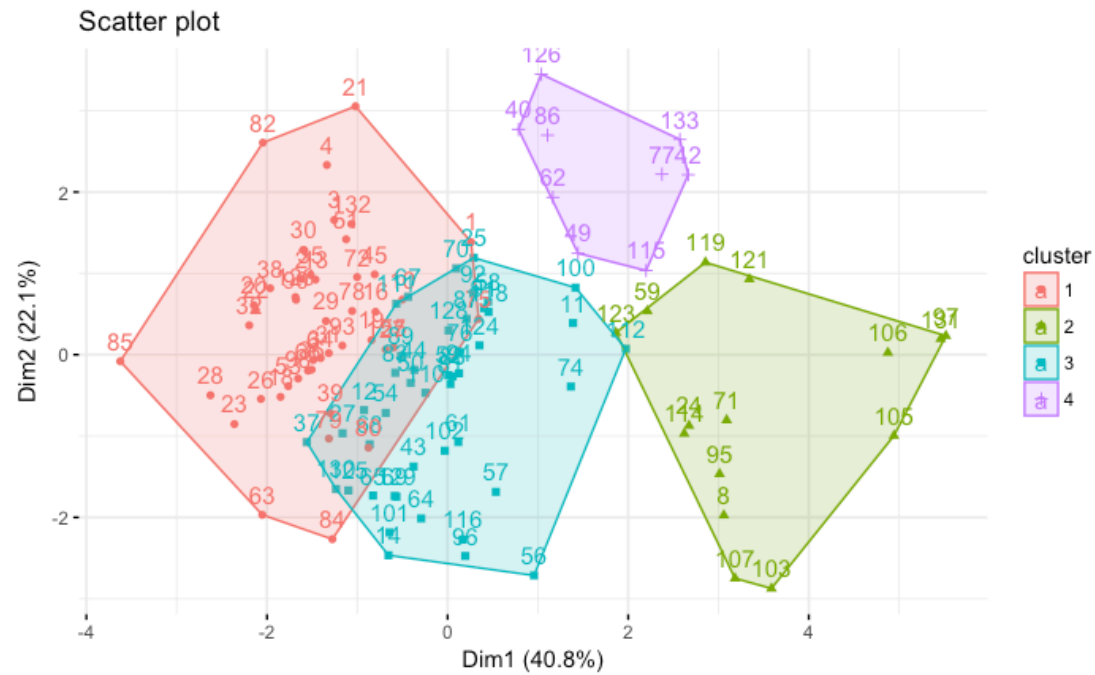


Figure 7.8. Visualisation of four-cluster solution, plotted using first two principal component dimensions. Each participant is represented by a data point and labelled with their participant number. Dimension 1 explains 40.8% of the variance, while dimension 2 explains a further 22.1%.

Due to the severity of their dysphagia, 22 participants did not complete the TWST and 29 participants did not complete the TOMASS. Results showed a multivariate main effect of cluster on the TWST outcomes, Pillai's trace = .30, $F(1, 90) = 12.64$, $p < .001$, and on the TOMASS outcomes, Pillai's trace = .51, $F(3, 81) = 4.05$, $p < .001$. Univariate analyses showed differences between the clusters for all the TWST variables (Table 7.8): volume/swallow [$F(1, 90) = 6.30$, $p = .01$], time/swallow [$F(1, 90) = 19.03$, $p < .001$], and volume/time [$F(1, 90) = 26.15$, $p < .001$], and the TOMASS variables: number of bites [$F(3, 81) = 3.26$, $p = .03$], number of masticatory cycles [$F(3, 81) = 13.79$, $p < .001$], and time [$F(3, 81) = 15.77$, $p < .001$]. No differences were found for number of swallows on the TOMASS [$F(3, 81) = 2.05$, $p = .11$].

Table 7.8

Comparison of TWST and TOMASS Outcomes by Cluster

		Cluster				<i>p</i>
Variable		1	2	3	4	
TWST	Vol/swallow	22.1 ± 9.3	12.3 ± 7.6 ^a	17.7 ± 9.8	14.0 ± 3.6	.01
	Time/swallow	1.7 ± 0.8	4.6 ± 2.9 ^b	3.4 ± 2.7 ^b	5.0 ± 3.2 ^b	<.001
	Vol/time	15.6 ± 8.7	4.2 ± 4.6 ^b	8.0 ± 6.1 ^b	4.1 ± 3.4 ^b	<.001
TOMASS	Bites	3.0 ± 1.4	4.6 ± 2.3 ^a	3.7 ± 1.5	3.6 ± 1.5	.03
	Masticatory cycles	56.8 ± 29.7	126.9 ± 49.5 ^b	84.7 ± 30.4 ^b	117.8 ± 53.3 ^b	<.001
	Swallows	2.5 ± 1.3	3.3 ± 1.5	3.6 ± 2.9	2.4 ± 1.3	.11
	Time	50.0 ± 24.3	117.3 ± 43.2 ^b	87.2 ± 33.7 ^b	114.9 ± 66.5 ^b	<.001

Note. Vol = volume, TWST = Timed Water Swallowing Test, TOMASS = Test of Masticating and Swallowing Solids.

^a Significantly different from cluster 1, $p < .05$.

^b Significantly different from cluster 1, $p < .01$.

Tukey post-hoc comparisons revealed that participants in cluster 1 had more efficient ingestion than the other clusters on both the TWST (Figure 7.9) and TOMASS (Figure 7.10). On the TWST, participants in cluster 1 were able to swallow a larger volume per swallow than cluster 2, and had a shorter time per swallow and larger swallowing capacity (volume/time) than clusters 2, 3, and 4. On the TOMASS, participants in cluster 1 required fewer bites of the cracker than cluster 2, and fewer masticatory cycles and less time to finish the cracker than clusters 2, 3, and 4. Cluster 2 had more masticatory cycles than cluster 3.

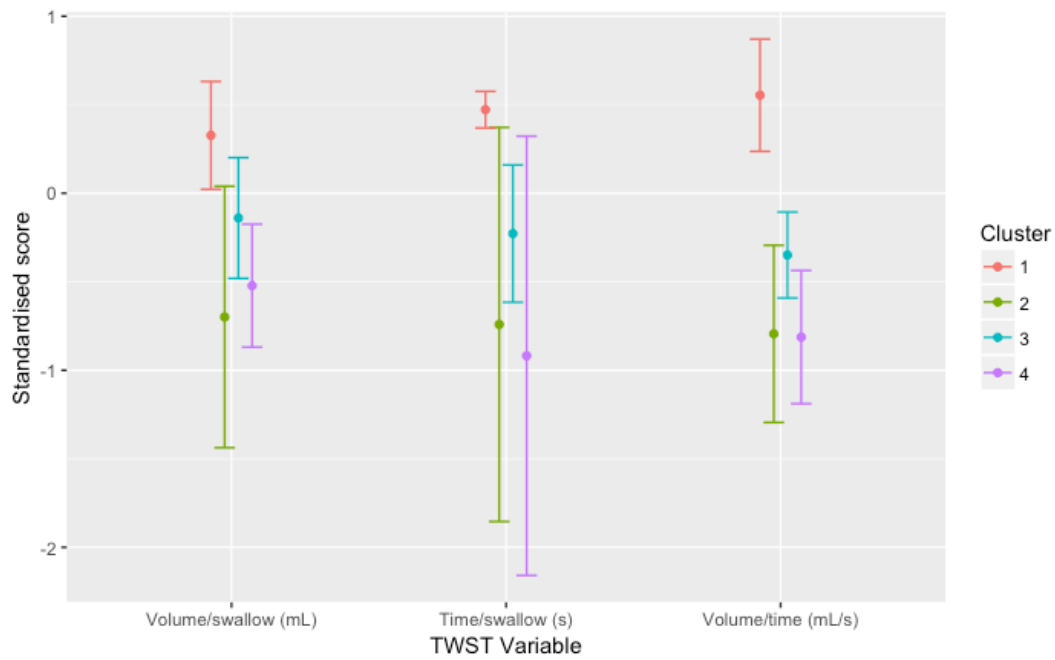


Figure 7.9. Timed Water Swallowing Test (TWST) standardised scores (mean and 95% confidence interval) for each cluster. The time per swallow scores have been reversed for ease of interpretation, so that a lower score reflects greater impairment.

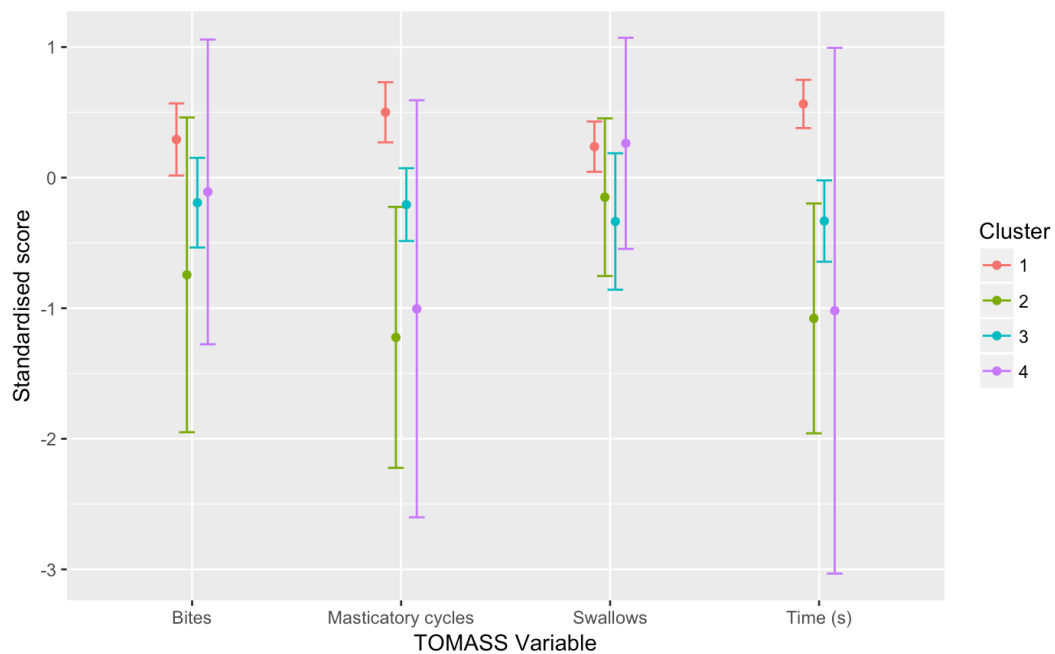


Figure 7.10. Test of Masticating and Swallowing Solids (TOMASS) standardised scores (mean and 95% confidence interval) for each cluster. Scores for all variables have been reversed for ease of interpretation, so that a lower score reflects greater impairment.

7.3.2.2 Variable reduction

There was a statistically significant strong negative correlation between jaw hit rate and jaw amplitude error ($r = -.81, p < .01$), as well as a moderate positive correlation between swallowing temporal error and jaw temporal error ($r = .64, p < .01$; Table 7.9). Swallowing hit rate had moderate negative correlations with both swallowing temporal error ($r = -.54, p < .01$) and swallowing amplitude error ($r = -.67, p < .01$).

Table 7.9

Correlation Coefficients (Spearman's rho) and P-Values for Strength and Movement Precision Variables

	ES	JF	SHR	STE	SAE	JHR	JTE	JAE
ES								
rho	1	0.24*	0.34**	-0.43**	-0.13	0.37**	-0.49**	-0.34**
p	0	0.01	0	0	0.16	0	0	0
JF								
rho	0.24*	1	0.03	-0.26**	0.02	0.24*	-0.36**	-0.26**
p	0.01	0	0.74	0	0.81	0.01	0	0
SHR								
rho	0.34**	0.03	1	-0.54**	-0.67**	0.13	-0.43**	-0.06
p	0	0.74	0	0	0	0.18	0	0.54
STE								
rho	-0.43**	-0.26**	-0.54**	1	0.08	-0.39**	0.64**	0.27**
p	0	0	0	0	0.39	0	0	0
SAE								
rho	-0.13	0.02	-0.67**	0.08	1	0.1	0.14	-0.08
p	0.16	0.81	0	0.39	0	0.3	0.13	0.38
JHR								
rho	0.37**	0.24*	0.13	-0.39**	0.1	1	-0.48**	-0.81**

p	0	0.01	0.18	0	0.3	0	0	0
JTE								
rho	-0.49**	-0.36**	-0.43**	0.64**	0.14	-0.48**	1	0.37**
p	0	0	0	0	0.13	0	0	0
JAE								
rho	-0.34**	-0.26**	-0.06	0.27**	-0.08	-0.81**	0.37**	1
p	0	0	0.54	0	0.38	0	0	0

Note. * $p < .05$. ** $p < .01$. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

The classification tree demonstrates that the most predictive variables for clustering participants were jaw-opening temporal error, jaw hit rate, and swallowing hit rate (Figure 7.11). The probability that the splitting rules were able to assign participants into the accurate clusters ranged from .73 – .89.

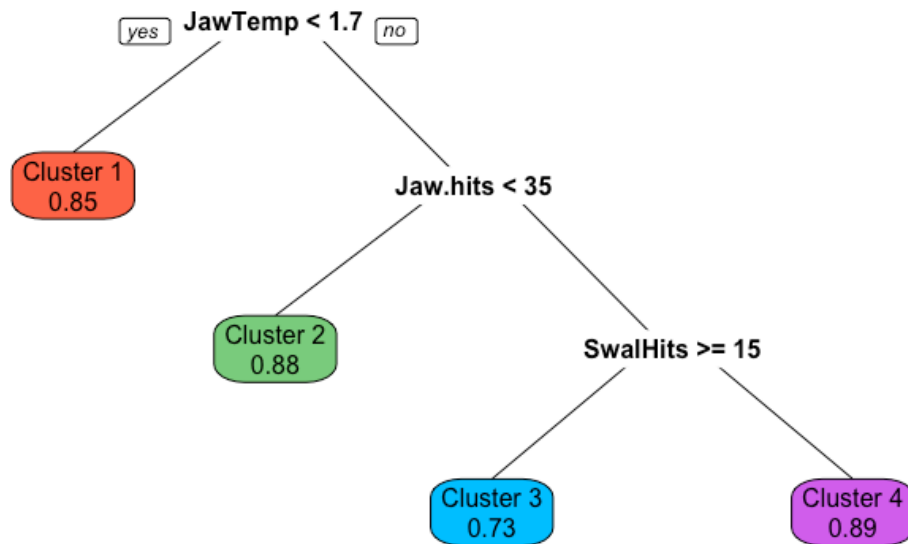


Figure 7.11. Classification tree for all participants, with the variables and cut-off scores that best predict their assignment into four clusters. Participants that match the splitting rule at the top of each split are assigned to the left branch. The numbers under the clusters indicate the

probability of the participants who match the splitting rule being correctly classed in that cluster.

A similar classification tree predicting cluster assignment was modelled for stroke patients only (Figure 7.12). The most predictive variables for stroke patients were jaw-opening hit rate, swallowing amplitude error, and jaw amplitude error. The probability that the splitting rules were able to assign patients into their accurate clusters ranged from .5 – 1.0.

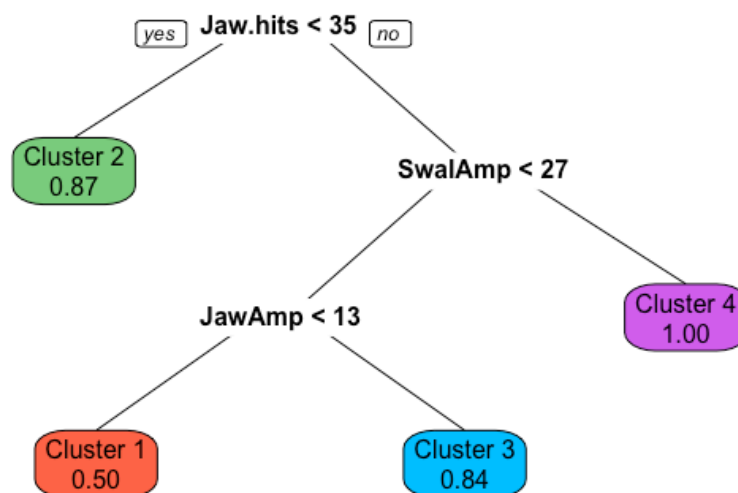


Figure 7.12. Classification tree for stroke patients only, with the variables and cut-off scores that best predict their assignment into four clusters. Participants that match the splitting rule at the top of each split are assigned to the left branch. The numbers under the clusters indicate the probability of being correctly classed in that cluster.

7.4 Discussion

It has historically been assumed that swallowing impairment seen after neurological damage is associated primarily with weakness as the underlying pathophysiology, even though reduced movement precision may contribute towards dysphagia. This assumption of weakness has been reinforced by the use of diagnostic methods that are limited to subjective inference as to the cause of dysphagia from visualised biomechanical impairments. A novel assessment protocol was developed to objectively measure and discriminate between strength and movement precision in swallowing. Based on test performance, there appeared to be several distinct

subgroups of dysphagia within the stroke patients. Measures of movement precision were found to be more sensitive than measures of strength in discriminating between clusters. The assessment was able to differentiate between diagnostic groups that were expected to have distinct patterns of strength and precision impairments, suggesting that the assessment holds promise in being a viable test of strength and movement precision. Assessment of movement precision impairment in swallowing may be an important but overlooked aspect of rehabilitation that should be further explored in controlled studies.

7.4.1 Classification of test performance using cluster analysis

A cluster analysis was used to explore underlying, previously undefined patterns in strength and movement precision among the participants, without using any known group labels such as diagnosis. The majority of healthy controls were assigned to a unique cluster (Cluster 1), and the patients with myopathy assigned to another distinct cluster (Cluster 3). This provides some internal validation of the cluster solution as it was able to discriminate between these two groups based only on performance. It was expected that stroke patients would be spread out across several different clusters because of the heterogeneity of swallowing impairments after stroke (Daniels & Huckabee, 2014; Perlman et al., 1994; Robbins et al., 1993). This expectation was met, as stroke patients were assigned to all four clusters. In addition, the stroke group's strength and movement precision performance values had much larger standard deviations compared to the healthy and myopathy groups, indicating greater variability within the stroke patients. This aligns with previous research indicating that stroke does not cause just one set of dysphagic symptoms, but instead results in a wide range of disorders (Daniels et al., 2006, 2009). However, caution should be used when interpreting the exploratory cluster analysis. Another measure of internal validity, the silhouette coefficient, suggested that the quality of cluster cohesion and separation was only poor to fair (Mooi & Sarstedt, 2011). This could be explained by the wide variability in patient performance. Clusters 1 and 3 had the lowest cohesion and separation scores, which may be due to the heterogeneous mix of healthy, stroke, and myopathy participants assigned to those clusters.

The performance of the clusters on the TWST and TOMASS provided some support for the external validity of the cluster solution. The cluster with adequate strength and movement precision (cluster 1) had significantly better scores on the TOMASS and TWST than the other three clusters that demonstrated impaired strength and/or precision, indicating that the assessment can separate those with and without functional swallowing deficits. However, using the TWST and TOMASS as an external validation measure of the cluster solution is limited,

since clinical tests of ingestion provide information about swallowing ability at a functional level, but are unable to characterise the underlying impairments of dysphagia (Hughes & Wiles, 1996).

Although participants in this study were predominantly assigned to the expected clusters, there was a minority which appeared to be misclassified. For example, while the majority of the healthy controls was assigned to cluster 1, six (15%) were placed in cluster 3 (characterised by decreased strength). These six participants could have demonstrated decreased strength measures because of normal sex- and age-related submental muscle force differences that do not affect functional swallowing. Since swallowing is a submaximal task, individuals can have a decrease in maximum strength and functional reserve but still maintain adequate force during swallowing (Nicosia et al., 2000). They were all female, with a higher mean age (75 years old) compared to the healthy group mean (69 years old). Research in healthy people has found that jaw-opening force is lower in females compared to men, and also lower in people with sarcopenia (Machida et al., 2017). None of the healthy participants were assigned to clusters 2 and 4 (clusters that demonstrated poor movement precision). This suggests that the clinical assessment had specificity for movement precision, so that people with intact movement precision were not assigned to the incorrect cluster. In addition, 16% of the stroke patients and 11% of the myopathy patients were assigned to cluster 1, demonstrating relatively intact strength and movement precision compared to the other patients with dysphagia. This could be explained by the possibility that their dysphagia was due to other underlying impairments (e.g., impaired sensation) that could not be identified by this assessment.

One interesting finding was in regard to the clusters' impairment patterns. Clusters characterised by relatively intact strength and movement precision, relative impairments in both strength and movement precision, and impairments in strength only were identified – however there was no evidence of a cluster with poor movement precision but intact strength. There could be several reasons for this absence. It could be that this pattern of impairment does exist, but was not present in our sample, or the assessment may not be sensitive to identifying patients with relatively poor movement precision but intact strength. Another explanation could be that, even though both strength and movement precision may contribute to functional swallowing, the ability to move precisely and accurately precedes the ability to generate maximum force. In this study, all patients with reduced movement precision also had decreased strength; in other words, none of the patients with decreased movement precision were able to score highly on the strength tests. The opposite was not true, since patients with muscle

weakness still had preserved movement precision and accuracy, within their decreased strength envelope. Perhaps there needs to be a certain amount of movement precision available in the submental muscles to generate enough muscle activity or force during swallowing-related behaviours.

Instead of a cluster with intact strength and poor movement precision, a pattern of poor strength, poor swallowing precision, yet relatively intact jaw movement precision emerged. Clusters 2 and 4, both made up almost entirely of stroke patients, had similarly impaired outcomes for strength and swallowing movement precision tasks. The only difference between these clusters of stroke patients was that cluster 4 had relatively intact accuracy of jaw movement, particularly amplitude precision, demonstrating that volitional jaw-opening precision can be intact while swallowing precision is impaired. This demonstrates the need for careful assessment of movement precision across different behaviours. Patients who present with intact precision for more voluntary movements (e.g., oral-motor, speech, and limb behaviours) may nevertheless have reduced swallowing movement precision which may be overlooked. Since volitional and reflexive swallowing behaviour are controlled by different cerebral regions (Doeltgen et al., 2011), there may also be task-dependent differences in the neural control of submental muscle function.

7.4.2 Variables predictive of cluster assignment

The cluster analysis assigned participants into four clusters based only on their performance on eight measures of strength and movement precision. Reducing the number of measures to those which are most efficient in determining cluster assignment can reduce the time needed to complete the assessment, improve productivity and cost-effectiveness, and reduce patient fatigue. The classification decision tree for all participants was able to reduce the number of salient test variables which predicted cluster assignment from eight to three. Only movement precision measures (jaw-opening temporal error, jaw-opening hit rate, and swallowing hit rate) were in the top three predictors, suggesting that movement precision measures might be a crucial part of diagnostic testing. Strength measures were not as effective in predicting cluster assignment. As can be seen in the graph depicting relative performance patterns (Figure 8.7), the maximum difference in strength test performance between clusters was approximately one standard deviation of the mean, while the maximum difference in movement precision performance was approximately 2.5 standard deviations. The larger differences in movement precision scores between clusters meant that movement precision tests were better able to discriminate between clusters.

A jaw-opening temporal error of less than 1.7% (equivalent to approximately 0.5 seconds) separated cluster 1, which had relatively intact strength and movement precision, from other clusters, which had impaired strength and/or decreased movement precision. This predictor was able to discriminate between cluster 1 and the other clusters with high accuracy (85%). Jaw-opening temporal precision might then be predictive of whether a patient has strength and/or movement precision impairments, and could potentially be used as a screening tool to identify those who warrant further testing. Both jaw-opening and swallowing hit rates were crucial predictors in assigning the rest of the participants to clusters 2, 3, and 4, indicating that movement precision of these behaviours can be differentially affected in people with dysphagia, and should be targeted in swallowing assessments.

Swallowing and jaw-opening hit rates had moderate-to-strong correlations with peak-to-target error values, suggesting that either hit rate or peak-to-target error measures could be removed from the assessment protocol in future research studies. Using hit rate as the sole measure of movement precision may be appropriate as it reflects both the temporal and amplitude error values. However, as it essentially is a binary hit/miss outcome measure, it is not a continuous measure and lacks the measurement precision with which the peak-to-target values are able to provide. An alternative measure that needs further investigation is the root mean square error between the response and the target (commonly used in other studies such as Van Hedel et al., 2010). Using this measure could reduce the number of variables that need to be analysed, and would take into account both temporal and spatial accuracy of the response.

7.4.3 Classification of test performance based on diagnostic groups

Since there is currently no gold standard measure of movement precision in swallowing for comparison, proof of concept for this assessment was achieved by comparing the performance of three diagnostic groups that were expected to have different patterns of impairment. As hypothesised, the novel assessment was sensitive to differences between healthy controls, stroke patients with dysphagia, and myopathic patients with dysphagia. Stroke patients with dysphagia had poorer performance on both strength and movement precision tasks compared to healthy controls, while myopathic patients with dysphagia had weakness with relatively intact movement precision. These patterns of impairment are consistent with previous research comparing ankle strength and skill between stroke and spinal cord injury patients (Van Hedel et al., 2010; Wirth et al., 2008). The assessment does not just simply discriminate between healthy controls without dysphagia, and patients with dysphagia. The fact that the two groups of patients with dysphagia also demonstrated different patterns of performance indicates that

the test can provide insight into the underlying mechanism of swallowing impairment due to different aetiologies. This may be useful for treatment planning.

The three groups were roughly matched in terms of sex and ethnicity. However, there were significant differences in age between the three groups, and the two patient groups also differed in the swallowing characteristics of dysphagia duration, diet level, and EAT-10 scores. Stroke patients were older, had more acute dysphagia, and were more likely to be on a modified diet, however they had less severe self-reported scores of swallowing impairment. The differences in acuity are to be expected given the development of dysphagia in an acute lesion such as stroke (Mann et al., 1999), compared to the chronic, progressive nature of myopathy (Oh et al., 2007; Oh, Brumfield, Hoskin, Kasperbauer, & Basford, 2008). Age affects jaw-opening force, with older individuals having decreased jaw force (Iida et al., 2013). Despite being older, the stroke patients still had higher jaw-opening force than the myopathy patients, but the difference might have been greater if the two groups were age-matched. Diet level and self-report of dysphagia symptoms on the EAT-10 can reflect dysphagia severity, but these measures can also be affected by patients' dietary preferences, dentition, and level of self-awareness regarding swallowing impairment. Despite the differences described above, objective and measurable scores of swallowing ability using the TOMASS and TWST were not significantly different between the dysphagia groups. Therefore, it is likely that dysphagia severity was not substantially different between stroke and myopathy groups. However, since patients with severe dysphagia or significant cognitive impairments were excluded from the study, the resulting stroke sample may have been biased towards those with mild-moderate impairments. If patients with severe dysphagia had been able to participate in the assessment, it is possible that the stroke group's performance would have been even more impaired in the strength tests, movement precision tests, or both, thus increasing the group differences seen in the current results.

7.4.4 Clinical implications

The clinical implications of these findings are important. Currently, decreased specificity of available assessment tools results in a lack of specificity in diagnosis; thus most dysphagic patients after stroke are assumed, by default, to be predominantly weak and prescribed strength training. However, based on the cluster profiles, this would describe and meet the needs of patients in cluster 3, which make up only 42% of the stroke sample. Another 41% of the stroke patients had decreased scores on the movement precision tasks, suggesting reduced coordination of timing and force in muscles used for swallowing. Using current methods of

assessment, these impairments would likely be misdiagnosed as weakness. Rather than the repetitive and progressive resistance exercises used in strength training, these patients might instead benefit from the complex, novel, and task-oriented aspects of skill training (Adkins et al., 2006; Athukorala et al., 2014). Further, as discussed above, it is possible that a certain level of movement precision is needed to generate adequate force during functional tasks. Thus, for patients with both strength and movement precision impairments, skill training may need to be initiated prior to strengthening exercises. Prescribing only strength training when a patient has movement precision deficits is not only an ineffective use of time and resources, but may cause unintended adverse consequences (Clark, 2003; Garcia et al., 2004). This preliminary evidence of movement precision impairment in dysphagic individuals suggests that accurate and specific diagnosis of swallowing pathophysiology is fundamental to the effective management of dysphagia, and that skill training is a logical and necessary target for swallowing rehabilitation (Huckabee & Kelly, 2006; Huckabee & Macrae, 2014).

7.4.5 *Future directions*

There are four issues related to this research that future investigations in this field should address. Firstly, a significant limitation is the lack of a gold standard assessment of skill impairment, without which makes it challenging to test the accuracy or validity of the strength and movement precision assessment. Significance testing could not be used to compare performance means between clusters, since the cluster solution was itself derived by maximising differences between clusters (Tan et al., 2005). Differences in strength and movement precision performance profiles of the clusters were compared relative to each other, and not to an external established norm. However, the exploratory nature of this study and the cluster analysis was expected, since this is the first time that strength and movement precision impairment has been measured and compared between different patient groups and healthy controls. Future work should focus on replicating and validating this cluster solution. External validation could be accomplished by replicating the current study with another sample of patients with dysphagia due to stroke, and comparing the actual with the predicted cluster solutions. Another method of validation could be to compare treatment outcomes between stroke patients who receive impairment-specific training, versus the alternative treatment. For example, patients who are found on the clinical assessment to have impaired movement precision (in relation to their strength) would be randomly assigned to impairment-specific training (in this case, skill training) or the alternative (strength training). The existence of subgroups within stroke-related dysphagia would be validated if the patients who participate in impairment-specific treatment have better outcomes than those who had the alternative.

A second limitation is that 13% of stroke patients were unable to participate in the clinical assessment due to fatigue, severe dysphagia, or cognitive deficits, resulting in a stroke sample that may be biased towards those with mild-moderate impairments. Anecdotally, patients reported having difficulty eliciting the 20 dry swallows needed for the clinical assessment within a single session, even when provided with rest breaks and sips of water as needed during and between tasks. The entire assessment session lasted an average of 1.5 hours, including obtaining informed consent and giving instructions, which was fatiguing for some patients. Instead of 5 – 10 trials for each task, perhaps three trials would be sufficient to capture individual variability while limiting patient fatigue. Future work should investigate the feasibility of using fewer trials per task, and should make efforts to recruit patients with different levels of severity.

Third, as previously discussed, the stroke, myopathy, and healthy groups were not age- and sex-matched, due to difficulty in recruiting adequate numbers of patients. The stroke group was significantly older than the healthy and myopathy groups, which may have contributed to the more severe strength and skill impairments demonstrated by the stroke patients. Future work should consider controlling for demographic and dysphagia differences between groups.

Finally, the reliability and precision of the novel assessment measures are unknown. It is vital that the measures of strength and movement precision are robust so that results of this study, and future studies, can be interpreted with confidence. If the assessment is to be used to identify strength and movement precision impairments, measurements need to be stable over time. Future studies should quantify the within-participant variation in the strength and movement precision measures, both within and across several sessions. In addition, the multiple measures of movement precision used in this study were found to correlate highly with each other, which may result in multicollinearity. The use of the root mean square value, which encapsulates both amplitude and temporal accuracy, may be considered as the sole measure of movement precision in future research. This value can be calculated by the BiSSkiT software and used as a quantitative measure of skill impairment during assessment, or provided to the patient and clinician immediately after the trial as biofeedback during training.

In conclusion, healthy controls, stroke patients, and myopathy patients participated in a novel assessment designed to quantify and discriminate between strength and movement precision impairment in swallowing. Several subtypes of swallowing pathophysiology were identified based on assessment performance, particularly in stroke patients. Results suggest that there can

be additional impairments in dysphagia other than weakness. However, this work is exploratory and requires replication and further evidence before it can be translated to the clinical realm. Movement precision measures appeared to be more predictive of cluster assignment than strength measures, suggesting that this is an important avenue for research into the pathophysiological features of dysphagia.

Chapter 8. Relationship between biomechanical measures of hyoid movement and physiological measures of strength and movement precision: An exploratory study (Study 3)

8.1 Introduction

Hyolaryngeal excursion during swallowing is important for airway protection (Logemann, 1998; Vandaele et al., 1995) and UES opening (Cook et al., 1989), thus contributing to swallowing safety and efficiency. Reduced hyoid displacement has been associated with many negative consequences, including increased risk of dysphagia (Perlman et al., 1994), aspiration (Bingjie et al., 2010; Steele et al., 2011), and pharyngeal residuals (Steele et al., 2011). In addition to spatial displacement, temporal aspects of hyoid bone movement such as duration and timeliness of initiation have also been found to be related to functional swallowing outcomes. Delayed initiation of hyoid bone elevation and prolonged duration of swallowing kinematics were associated with the presence of aspiration in stroke patients (Bingjie et al., 2010). Measures of velocity can also provide information about the rate of hyoid bone movement, combining both spatial and temporal aspects of movement (Sia et al., 2015). Reduced hyoid velocity was demonstrated in patients with dysphagia secondary to stroke (Seo, Oh, & Han, 2016) and nasopharyngeal carcinoma (Wang, Chang, Chen, Lin, & Hsiao, 2010) when compared to healthy controls; further, the patients who aspirated had slower velocity than patients who did not aspirate (Wang et al., 2010).

Given the relationship between abnormal hyolaryngeal kinematics and dysphagia, studies have attempted to investigate the mechanisms causing these abnormalities. Contraction of the submental muscle group contributes towards movement of the hyolaryngeal complex (Pearson et al., 2013), and thus it would be expected that decreased strength of the submental muscles might be related to impaired hyoid biomechanics, such as decreased range of motion. However, research investigating this relationship has been inconclusive. Since the submental muscles are also responsible for opening the jaw, strength of the submental muscles can be assessed during maximum jaw-opening against resistance, using a dynamometer fixed under the chin. It was found that reduced submental muscle strength (as measured using jaw-opening force) was correlated with increased hyoid displacement in healthy elderly men (Shinozaki et al., 2017). The study also reported that decreased jaw force was related to a low resting hyoid position, but was not significantly correlated with maximum hyoid position. Nor were there associations between jaw force and any measures of hyoid displacement or position in healthy elderly female participants (Shinozaki et al., 2017). However in another study, patients with myopathy

had reduced hyoid excursion and velocity, demonstrating a possible relationship between muscle weakness and reduced hyoid movement (Paik et al., 2008). It is unknown whether measures of strength, such as jaw-opening force and peak sEMG amplitude during effortful swallowing, are related to hyoid movement for other patient groups with dysphagia. In addition, the direction of this relationship remains unclear.

Historically, it has been assumed that impaired hyoid biomechanics visualised on VFSS were caused primarily by the pathophysiology of weakness. However lesions of the central nervous system, such as from stroke, can cause numerous physiological impairments in corticobulbar muscle function other than strength, including but not limited to abnormalities in precision, tone, and motor planning (Duffy, 2005). Deficits of strength and movement precision after stroke may both cause slowed or reduced amplitude of hyoid movement, and may look the same at a biomechanical level. For example, reduced hyoid range of motion may be caused by decreased muscle activation and poor force generation (weakness), resulting in inadequate degree of maximal hyoid displacement from rest. On the other hand, decreased temporal and spatial coordination of submental muscle contraction can result in poor accuracy and precision of hyoid bone movement to reach an optimal maximum position. No research has investigated the relationship between impaired hyoid movement on VFSS and decreased movement precision. Stroke patients with suspected swallowing apraxia and incoordination of the oral musculature demonstrate a delay in initiation of bolus transfer in the oral stage of swallowing (Daniels, 2000). It is possible that a patient with decreased temporal accuracy during a movement precision test might also manifest a delay in initiation or duration of hyoid movement in the pharyngeal phase. In the limb literature, decreases in both muscle strength and coordination contribute towards biomechanical impairments such as gait speed (Tomita & Usuda, 2013). Abnormal movement of the hyoid bone, therefore, might be associated with impairments in strength, movement precision, or both.

The aim of this study was to explore the relationship between biomechanical movement of the hyoid bone seen on VFSS, with performance on physiological measures of strength and movement precision in stroke patients. It was expected that stroke patients with different patterns of strength and movement precision deficits would also differ on VFSS kinematic measures of hyoid movement, thus demonstrating a relationship between physiological and biomechanical measures of hyoid movement. This exploratory study will provide evidence about the nature of these impairments underlying dysphagia after stroke, supporting further research in the area of stroke rehabilitation.

8.2 Methodology

8.2.1 *Participants*

The performance of stroke patients on the strength and movement precision assessment from Study 2 were compared to the available normative data collected from 35 healthy participants. Normative data were stratified by age (50 – 69 years and 70+ years) and sex. Reduced movement precision was defined as having swallowing hit rate, amplitude error, and temporal error worse than age- and sex-matched healthy adults (i.e., swallowing hit rate below the lower 95% confidence interval (CI) of the mean, and amplitude and temporal errors higher than the upper CI). Reduced strength was defined as jaw-opening force below the lower CI of the mean for age- and sex-matched healthy adults. If patients met the criteria for having reduced movement precision but adequate strength, or having reduced strength and adequate movement precision, they were asked to participate in this study (Study 3), which consisted of a VFSS within two weeks of participation in Study 2. Sample size calculations suggested that a sample size of eight (four in each group) was needed. However, it was difficult to find adequate numbers of patients to participate in a VFSS, as many were hospitalised and unable to travel to the research laboratory. In addition, it was found that most stroke patients did not meet the inclusion criteria of having impairments in only the strength tests or only the movement precision tests, and instead demonstrated reduced performance in at least one strength test as well as one movement precision test. Therefore, approximately one month into the year-long study, it was decided that from that time forward every stroke patient who participated in Study 2 would also be asked to take part in Study 3, regardless of their test performance. Instead of comparing VFSS measures between two stroke groups, the relationship between physiological and biomechanical measures for all patients would be explored using correlation coefficients. Written informed consent was obtained prior to data collection, and the study was approved by the appropriate regional Human Ethics Committee (see Appendices).

8.2.2 *Experimental procedure*

VFSS were completed using a Philips C-arm fluoroscope at 25 frames/second in the lateral view, with recordings captured on the Kay Elemetrics Digital Swallowing Workstation (KayPentax, Lincoln Park, NJ, USA). Participants were seated comfortably in an upright position. A radiopaque disk (19 mm diameter) was placed on the participant's anterior neck using medical adhesive tape to allow for calibration of displacement measures during subsequent data analysis. The field of view included the lips and cervical spine anteriorly and posteriorly, and the oral cavity and cervical oesophagus superiorly and inferiorly. Participants

were presented with three trials of 5 mL thin liquid barium via cup sip, and then three trials of 5 mL puree via teaspoon (40% w/v ratio of barium sulphate concentration; Varibar Barium Sulfate Contrast, Thin liquid and Pudding, E-Z-EM Canada Inc). It is recommended that at least three trials of each volume/consistency be completed during VFSS “in an effort to balance the need to capture the individual variability with the negative effects of radiation exposure” (Lof & Robbins, 1990, p. 242). Participants were instructed to hold the bolus briefly in their mouth, before a verbal command for a cued swallow.

8.2.3 Outcome measures

VFSS recordings of each swallowing trial were transferred to a personal computer as separate video segments. Two video segments could not be analysed: the cervical spine moved out of view during a puree trial for Participant 45, and the recording of one liquid trial for Participant 120 was accidentally deleted. There were 41 single-swallow video segments in the final analysis. QuickTime Player software (Version 10.4, Apple Inc) was used to advance and reverse through each recording frame-by-frame to identify the frame number for the following biomechanical events: (a) bolus “hold” position (bolus in anterior oral cavity, just before any posterior movement), (b) bolus head reaching ramus of mandible, (c) onset of hyoid burst movement, and (d) time of maximum hyoid displacement. Temporal measures (in seconds) were calculated by subtracting frame (c) from (d) to determine duration of hyoid burst movement, and subtracting (c) from (b) to determine STD, and multiplying number of frames by 1/25. STD is a measure of time between the end of the oral phase (marked by bolus head reaching ramus of the mandible) and the beginning of the pharyngeal phase, with prolonged durations suggesting a delay in pharyngeal initiation. When reduced glossopharyngeal approximation resulted in pre-swallow pooling of the bolus in the pharynx, STD onset was defined as the frame where the base of tongue dropped during volitional oral transfer.

Spatial measurements were completed using image processing software (ImageJ, National Institutes of Health, Bethesda, MD). The bolus hold and maximum hyoid displacement frames were copy and pasted from QuickTime Player into ImageJ. The y-axis was aligned with the spine, by drawing a line between the anterior inferior corners of two cervical vertebrae (C2 or C3, to C4) to serve as the y-axis, and drawing the x-axis as a straight line perpendicular to the y-axis and originating from the anterior inferior corner of C4. The images were calibrated to the radiopaque disk, and the anterior superior corner of the hyoid was traced on each frame. The maximum hyoid displacement frame was then copy and pasted on top of the bolus hold frame so that the x- and y-axes of the two frames were aligned (frames were rotated as needed).

A line was drawn between the hyoid tracing at rest and at maximum displacement, with maximum hyoid displacement measured as the length of this line in mm. Velocity of hyoid burst (in mm/s) was calculated by dividing maximum hyoid displacement by duration of hyoid burst.

8.2.4 *Statistical analysis*

A multivariate analysis of variance (MANOVA) was run to investigate trial effect. Trial was included as the independent variable, while the eight VFSS hyoid measurements were included as dependent variables. The relationship between the eight VFSS variables from this study, and the eight strength and precision variables from Study 2, was analysed using Spearman rank correlations. Significance values of the correlation coefficients were calculated using permutation methods to control for multiple comparisons.

8.3 Results

Eight stroke patients participated in this study. One participant's data was not used because the hyoid could not be viewed on the videofluoroscopy recording, due to overexposure of the image. Due to difficulty recruiting adequate numbers of stroke patients who were able to participate in a VFSS, another patient was not recruited. Data analyses were completed for the remaining seven patients. Patient demographics and their results from the clinical strength and movement precision assessment are presented in Table 8.1.

Table 8.1

Participant Demographics and Results from Strength and Movement Precision Assessment

Participant	Age	Sex	Cluster	ES	JF	SHR	STE	SAE	JHR	JTE	JAE
8	69	F	2	0.9	36.1	30.0	19.4	4.8	20.0	2.6	26.1
42	79	M	4	1.2	57.2	0.0	16.9	29.7	50.0	5.1	14.1
45	68	M	1	1.9	84.9	40.0	2.1	23.0	70.0	1.9	11.4
49	74	F	4	0.8	66.4	10.0	10.4	19.2	60.0	4.9	11.9
111	67	M	3	1.7	77.0	40.0	4.9	17.8	70.0	2.2	10.0
120	66	M	1	1.2	157.6	50.0	1.7	16.7	70.0	1.1	10.8
124	70	M	3	1.1	69.5	30.0	11.9	10.6	60.0	1.9	10.8

Note. ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

There was no main effect of trial on VFSS measurements [Pillai's trace = .66, $F(2, 16) = 0.61$, $p = .84$], so each participant's VFSS measurements for each condition were averaged over three trials. Measurements of hyoid kinematics from the VFSS are shown in Table 8.2.

Table 8.2

Means \pm Standard Deviations of Hyoid Kinematic Measurements on VFSS

Patient	Displacement (cm)		Duration (s)		Velocity (cm/s)		STD (s)	
	Liquid	Puree	Liquid	Puree	Liquid	Puree	Liquid	Puree
8	1.6 \pm 1.1	1.5 \pm 0.6	0.4 \pm 0.1	0.6 \pm 0.1	4.1 \pm 2.6	2.4 \pm 1.1	1.3 \pm 1.8	-0.2 \pm 0.4
42	2.0 \pm 0.3	1.6 \pm 0.1	0.4 \pm 0.1	0.7 \pm 0.1	5.5 \pm 0.4	2.3 \pm 0.2	0.6 \pm 0.7	8.3 \pm 0.7
45	3.0 \pm 1.0	2.2 \pm 0.4	0.6 \pm 0.3	0.7 \pm 0.4	5.7 \pm 3.4	5.0 \pm 0.8	-0.1 \pm 0.2	0.4 \pm 0.9
49	1.8 \pm 0.5	1.9 \pm 0.2	0.3 \pm 0.0	0.4 \pm 0.1	5.3 \pm 1.6	5.3 \pm 1.6	0.8 \pm 0.1	2.7 \pm 2.5
111	1.6 \pm 0.2	1.8 \pm 0.2	0.4 \pm 0.1	0.7 \pm 0.3	4.4 \pm 0.3	3.0 \pm 1.1	-0.1 \pm 0.2	1.0 \pm 0.3
120	1.3 \pm 1.1	1.9 \pm 0.2	0.2 \pm 0.2	0.6 \pm 0.2	5.8 \pm 3.8	3.2 \pm 1.0	0.0 \pm 0.1	1.3 \pm 0.4
124	1.7 \pm 0.3	1.9 \pm 1.8	0.4 \pm 0.0	0.3 \pm 0.3	4.8 \pm 0.9	7.1 \pm 7.2	0.3 \pm 0.2	0.3 \pm 0.3
Mean	2.0 \pm 0.7	1.8 \pm 0.4	0.4 \pm 0.2	0.6 \pm 0.2	5.0 \pm 1.7	4.0 \pm 2.1	0.5 \pm 0.8	2.0 \pm 2.9

Note. STD = stage transition duration.

Four of the eight variables (hyoid displacement for liquid bolus, hyoid burst duration for liquid and puree, and STD for puree) and all the eight strength and movement precision assessment variables had non-normal distributions, as measured by the Shapiro-Wilk test. Thus, non-parametric Spearman rank correlations were completed to assess the strength and direction of the relationship between the eight clinical measurements of strength and precision and the eight VFSS measurements of hyoid movement (Table 8.3). There was a strong positive correlation between hyoid burst duration for a puree bolus and effortful swallowing ($r = .83$, $p = .03$). Stage transition duration for liquid was negatively correlated with effortful swallowing ($r = -.93$, $p = .01$), jaw force ($r = -.86$, $p = .02$), and jaw hit rate ($r = -.90$, $p = .02$), and positively correlated with swallowing temporal error ($r = .79$, $p = .05$).

Table 8.3

Spearman Correlation Coefficients and Significance Values for the Relationship Between Clinical and VFSS Measures

VFSS measures	Clinical measures							
	ES	JF	SHR	STE	SAE	JHR	JTE	JAE
Disp. – Liquid								
rho	0.50	0.61	0.24	-0.64	0.64	0.51	-0.25	-0.14
<i>p</i>	0.27	0.17	0.60	0.14	0.14	0.25	0.59	0.78
Disp. – Puree								
rho	0.43	0.75	0.45	-0.68	0.14	0.67	-0.68	-0.50
<i>p</i>	0.35	0.07	0.30	0.11	0.78	0.12	0.10	0.27
Duration – Liquid								
rho	0.63	0.13	0.44	-0.09	0	0.17	-0.11	0.07
<i>p</i>	0.14	0.80	0.33	0.86	1.00	0.73	0.81	0.88
Duration – Puree								
rho	0.83*	0.34	0.30	-0.38	0.63	0.43	0.09	-0.13
<i>p</i>	0.03	0.46	0.51	0.40	0.15	0.32	0.85	0.79
Velocity – Liquid								
rho	0.43	0.64	0.29	-0.68	0.54	0.51	-0.32	-0.21
<i>p</i>	0.35	0.14	0.54	0.11	0.23	0.25	0.49	0.66
Velocity – Puree								
rho	-0.14	0.39	0.16	-0.36	-0.18	0.36	-0.50	-0.36
<i>p</i>	0.78	0.39	0.73	0.44	0.72	0.43	0.26	0.45
STD – Liquid								
rho	-0.93**	-0.86*	-0.69	0.79*	-0.32	-0.90*	0.57	0.75
<i>p</i>	0.01	0.02	0.10	0.049	0.50	0.02	0.20	0.07
STD – Puree								
rho	0	0.07	-0.33	-0.21	0.75	0.11	0.43	-0.04
<i>p</i>	1.00	0.91	0.48	0.66	0.07	0.82	0.35	0.96

Note. * $p < .05$, ** $p < .01$. Disp. = displacement, STD = stage transition duration, ES = Effortful swallowing, JF = Jaw force, SHR = Swallowing hit rate, SAE = Swallowing amplitude error, STE = Swallowing temporal error, JAE = Jaw amplitude error, JTE = Jaw temporal error.

8.4 Discussion

This research represents first steps in investigating relationships between novel physiological measures of strength and movement precision, and biomechanical dimensions on VFSS. As submental muscles are assumed to be important for hyolaryngeal excursion, one might expect that there would be correlations between physiological measures of submental strength and movement precision, and biomechanical measures of hyoid movement. For example, a measure of submental strength such as jaw-opening force might correlate with hyoid displacement or velocity during swallowing, and temporal accuracy measured with submental sEMG in the movement-precision test might be related to temporal measures of hyoid movement duration or velocity. This was based on previous findings demonstrating a relationship between decreased strength and impaired hyoid movement (Paik et al., 2008; Shinozaki et al., 2017), and an association between temporal incoordination in the ankle and abnormal gait biomechanics (Tomita & Usuda, 2013). This study was exploratory in that there has been no previous research on swallowing movement precision in the submental muscles and its biomechanical correlates. Results from this study demonstrated that overall, there were few significant relationships between physiological measures and biomechanical hyoid movement. Hyoid trajectory was not specifically associated with measures of swallowing-related submental strength nor movement precision. Given the preliminary nature of this study, further research in the area is warranted.

There are several possible explanations for these negative results. Motor impairment in the limbs after stroke may be caused by several mechanisms, including but not limited to decreased muscle activation to generate force, impaired precision, muscle overactivity, and impaired motor planning (Gracies, 2005a). In swallowing, the underlying pathophysiology of dysphagia after stroke is not well-defined, but hyoid movement could be influenced by multiple pathophysiological causes, similar to the limbs. In this study, only measures of strength and movement precision were investigated. The limited number of strong correlations between the physiological and biomechanical measures could be explained by the possibility that hyoid movement in these patients was more affected by pathophysiological causes other than strength and movement precision.

Even though VFSS is considered the “gold standard” of dysphagia assessment and can provide a clear visualisation of biomechanical movement (Kendall & Leonard, 2001; Nagy, Molfenter, Péladeau-Pigeon, Stokely, & Steele, 2014), the specific motor impairment underlying the abnormal mechanical movement can only be inferred, and not directly measured, from VFSS

(Huckabee & Macrae, 2014). In the limb literature, biomechanical measures which assess motor performance of the arm are not challenging enough to truly measure strength (Bohannon, 2007). Patients who were judged as having intact arm functioning, based on biomechanical measures, had less than 50% of expected arm strength when measured with a dynamometer. Therefore, spatial and temporal measures of structural movement can provide a general indication of muscle function, but do not specifically assess the ability of muscles to generate force. This is consistent with findings that the maximum hyoid position on VFSS in healthy elderly men and women does not correlate with submental muscle strength measured using jaw-opening force (Shinozaki et al., 2017). Further, while VFSS may be able to detect skill impairment at a functional level (i.e., decreased swallowing safety), it is likely unable to quantify the level of impairment in movement precision, as it does not specifically assess the accuracy and precision of a goal-directed behaviour.

Finally, another explanation for the results could be that our definition of movement precision needs refining, and that both VFSS and the clinical assessment did not fully capture the complexities of movement precision in swallowing. Given the novelty of the clinical assessment, and the lack of previous research on movement precision in swallowing, our measurement of movement precision as the temporal and spatial accuracy of hitting the sEMG target on BiSSkiT has not yet been proven to be reliable nor valid. Some researchers have defined skilled movement as the ability not just to move accurately, but to also move with adequate speed (Reis et al., 2009; Shabbott et al., 2013). Speed and accuracy are inter-related in a concept termed the speed-accuracy trade-off, which states that more accurate movements are performed at the cost of decreased speed, while increasing movement speed results in a loss of accuracy (Fitts, 1964). Quantifying skill requires the concurrent assessment of both accuracy and speed (Willingham, 1998). The speed-accuracy trade-off might be present in the swallowing mechanism, as evidenced by increased swallowing velocity being associated with less efficient swallowing behaviour (e.g., increased oral and pharyngeal residuals; Pauloski et al., 2009). The poor accuracy demonstrated by stroke patients in this study could have reflected increased movement speed, instead of impaired movement precision. However, the lack of association between accuracy measures on BiSSkiT and hyoid velocity on VFSS does not support this idea. Further research should elucidate whether the concept of a speed-accuracy trade-off, as seen in skilled limb movement, applies to a complex, patterned behaviour such as swallowing.

While there were only five significant correlations out of the 64 pairs of variables, four of the five significant correlations involved STD. There was a reduction in STD as swallowing temporal error decreased and jaw hit rate increased. This suggests that the timely initiation of hyoid movement and pharyngeal swallowing was associated with improved movement precision. A shorter STD was also significantly associated with increased effortful swallowing amplitude and jaw opening force. This finding was interesting because it was not anticipated that STD, a measure traditionally associated with oropharyngeal sensation (Power et al., 2007) and temporal coordination (Logemann, 1998), would be correlated with measures of strength. There was a wide variability of STD values in this study, which has also been noted in previous research (Molfenter & Steele, 2012). Results from this study suggest that the cause of delayed hyoid initiation may be multifactorial, warranting further investigation.

Limitations of this exploratory study must be addressed. Due to a small sample size of seven patients, the study might not have had adequate statistical power to detect a true effect. It was difficult to recruit a larger sample of patients who could travel to a facility with VFSS. Future studies could consider using an assessment method that can measure hyoid kinematics at bedside, for example, ultrasonography. This might increase the sample size. In addition, it is unknown whether the physiological and biomechanical measurements were taken on the exact same behaviour in each patient. A more accurate representation of the relationship between physiological and kinematic measurements might be obtained if the assessments were completed simultaneously, i.e., the strength and movement precision assessment conducted under fluoroscopy. Finally, it is important to note that correlation does not equal causation, and any associations might have been confounded by factors which are known to affect hyoid kinematics, such as sex, age, and location of stroke lesion (Kendall & Leonard, 2001; May et al., 2017; Molfenter & Steele, 2013).

In conclusion, this study represents an initial step in exploring the pathophysiology underlying abnormal hyoid bone movement. There were limited significant relationships between measures of strength, movement precision, and hyoid kinematics. The results could be explained by the possibility that 1) strength and movement precision did not affect hyoid movement in this sample, and there were other impairments causing abnormal biomechanics, 2) VFSS lacked the ability to adequately and directly measure underlying pathophysiology, and 3) our definition and measurement of movement precision in swallowing was not valid. Further research is needed to refine measures of movement precision before the relationship between impairment in submental muscle contraction and biomechanical movement of the

hyoid can be fully investigated. Better understanding of the pathophysiology causing impaired hyoid bone movement will lead to improved rehabilitation techniques for patients after stroke.

Chapter 9. Conclusion

This research programme represents an initial foray into the development of a clinical test to identify strength and movement precision deficits underlying dysphagia. Since the minimum sEMG threshold during swallowing has now been established at 30% of maximal swallowing contraction, the skill-based biofeedback software can provide targets for swallowing execution that are within a physiologically-achievable range for both magnitude and timing. Preliminary data indicate that performance on the assessments of strength and movement precision was related to aetiology of dysphagia and functional swallowing outcomes, but not biomechanical abnormalities. In particular, the movement-precision measures of hit rate and spatiotemporal error during swallowing and jaw-opening were more strongly able to predict the pattern of swallowing impairments exhibited by participants than strength measures could. Therefore, the novel assessment demonstrates potential to differentially diagnose two possible pathophysiologies underlying dysphagia, as an adjunct tool to current diagnostic methods, and warrants further investigation. The assessment is non-invasive, relatively inexpensive and portable, allowing patients to be tested in the clinic, hospital, or community. The majority of healthy older participants and patients with dysphagia due to central and peripheral lesions were able to participate in testing without difficulty. However, future research should further refine the validity, reliability, and precision of the assessment measures before the assessment can be clinically translated.

Previous research has demonstrated the importance of identifying the pathophysiological mechanisms underlying dysphagia and matching these impaired mechanisms with specific rehabilitation techniques. This thesis contributes to the growing literature by identifying the presence of dysphagia subgroups in stroke patients and providing characterisation of the impairment patterns of each subgroup. Results reinforce the emerging notion that strength deficits are not always the primary contributor to swallowing impairment, and suggest that there may be other impairments causing dysphagia, including (but not limited to) movement precision deficits. Impairments in movement precision were defined, in this research programme, as decreased accuracy in the spatial and temporal aspects of movement in relation to environmental needs during swallowing. The possible presence of movement-precision impairments, particularly in patients with central nervous system damage, has implications on the choice of treatment techniques for patients with dysphagia. Currently, the majority of traditional therapy techniques prescribed to patients are strength-based exercises (Macrae & Humbert, 2013). However, results from Study 1 support previous research demonstrating that

functional swallowing is submaximal, and provides evidence that it is more similar to minimum effort swallowing. This questions whether there is ever a need for intensive maximum-effort exercises in swallowing rehabilitation. Strengthening exercises, such as head lifts (Shaker et al., 2002) and EMST (Troche et al., 2010), are designed to increase submental force generation and hyoid displacement, and are recommended for rehabilitation of patients with presumed weakness. However, results in Study 3 failed to confirm a relationship between decreased submental muscle strength and abnormal hyoid movement in patients with dysphagia, and suggest that the contribution of weakness to biomechanical impairment may be less significant than previously thought. In Study 2, a proportion of patients with dysphagia after stroke performed poorly on the strength tests (similar to patients with myopathy), but a similar proportion had impairments on the movement precision tests in addition to weakness. Strength training may still be indicated to target decreased strength. However, for stroke patients with deficits in movement precision, the functional repetition of submaximal targets in skill-based training (Athukorala et al., 2014) may be a valuable therapeutic tool.

Clinicians and researchers should be mindful of the importance of specificity in dysphagia assessment. Deficits in movement precision may be different in voluntary versus swallowing behaviours, and may be unrelated to an individual's strength level. The evaluation of movement precision should be completed separately from strength, and across different behaviours. The current gold standard evaluation, VFSS, is undoubtedly a valuable technique for visualising the swallowing response as a whole, illustrating bolus flow, and identifying structural movement abnormalities in the oral, pharyngeal, and cervical oesophageal phases. However, VFSS cannot quantify the magnitude or precision of muscle contraction underlying biomechanical movement. Alternative assessments can provide unique and complementary information about the pathophysiology of dysphagia. When stabilised to the chin and head, dynamometry can measure maximum voluntary contraction of jaw-opening against resistance, providing a measure of force generation and strength. sEMG biofeedback provides a simple interface which measures the temporal accuracy and precise gradation of muscle contraction during goal-directed behaviour. It is possible that each assessment modality is specialised at measuring a particular aspect of swallowing impairment, and multiple diagnostic tools may be needed to glean a full picture of the patient's multifaceted swallowing problems. Therefore, there is a need for further research to develop accurate, objective measures of the different pathophysiological features of swallowing, that will translate to improved diagnosis and treatment of dysphagia.

The emergence of skill training in dysphagia signals a shift from peripheral strength-based exercises to centrally-mediated techniques. Although some of these emerging skill-based exercises have demonstrated physiological and functional changes in patients after treatment (Athukorala et al., 2014; Steele et al., 2013; Stepp et al., 2011), it is still unknown how to identify those patients which would benefit from skill or strength training. Research studies investigating skill training in dysphagia have enrolled patients of a certain aetiology (e.g., Parkinson's or stroke), but not controlled for the underlying pathophysiology (e.g., reduced movement precision) as part of inclusion criteria (Athukorala et al., 2014; Stepp et al., 2011). This likely results in a heterogeneous sample. As demonstrated in this research programme with stroke patients, patients within the same diagnostic group may have different patterns of strength and movement precision impairments. Patients with heterogeneous impairments may react differently to treatment, increasing variability within the sample. Studies of skill and strength training have been completed before a full understanding of the factors affecting underlying impairment after stroke has been established.

It would be premature to come to any conclusions regarding the multi-factorial mechanisms of weakness and movement precision in swallowing from results of these exploratory studies, however some speculations can be made. The nature of weakness may be different depending on central versus peripheral lesions, with upper motor neuron damage and secondary muscular adaptations to denervation causing weakness after stroke, and peripheral muscle atrophy resulting in weakness in myopathic disease (Briani et al., 2006; Ng & Shepherd, 2000; Turner & Hilton-Jones, 2010). Movement precision in swallowing is likely associated with cortical control, since poor performance on the movement precision tests was seen mainly in stroke patients and not the other groups. Disruption of descending cortical pathways causing abnormal modulation and synchronisation of motor unit firing rates may be one mechanism causing decreased spatiotemporal accuracy and impaired precision of movement (Canning et al., 2000; Semmler, Sale, Meyer, & Nordstrom, 2004). Considerably more work is required to determine the extent to which deficits in strength and movement precision affect the safety, efficiency, and enjoyment of swallowing. Future work should focus on investigating the relevance and nature of strength and movement precision deficits in dysphagia after stroke, which will provide stronger justification for strength- and skill-based rehabilitation protocols. The work contained in this thesis provides a starting point for future discussion and research.

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Appendices



Participant Information Sheet

Effect of effort and bolus type on submental surface electromyographic activity during swallowing

I am a PhD student completing my doctoral degree in the Department of Communication Disorders, under the supervision of Prof Maggie-Lee Huckabee. You are invited to take part in a study measuring swallowing skill in healthy adults from 4 age groups – 18-35, 35-50, 50-65, and 65-90 years old. The purpose of this study is to see if muscle activity in the swallowing muscles changes when people swallow with maximum, normal, or minimal effort, and if age affects this skill. Results from this study will give us information on how to design assessment and treatment programs for patients with swallowing problems.

You can join this study if you are between the ages of 18-90 years, and have no medical problems that may affect your swallowing. If you choose to take part in this study, your involvement in this project will be to swallow your saliva and 5 mL sips of water. You will be asked to swallow in 3 different ways – swallow as hard as you can, swallow normally, and swallow as softly as you can. In total you will be asked to swallow 30 times, every 12 seconds. The muscle activity of your muscles used for swallowing will be measured using an electrode attached to the skin under your chin. Participants will need to be clean-shaven to allow good electrode-to-skin contact. The electrode can be easily removed after the study is completed. The entire session will take approximately 30 minutes. There are no known risks of using small sensors to monitor muscle activity, formally known as surface electromyography.

Participation is voluntary and you have the right to withdraw at any stage without penalty and without having to give a reason. This will in no way affect any future care or treatment, or academic participation if you are a student. If you withdraw after completing the session, you cannot remove your data, as we will remove your name and identifying information at that point in time.

Measurements of your swallowing will be stored on a computer and analysed at a later time. The only data recorded will be the line tracings that represent your

swallowing, and your date of birth. To ensure anonymity and confidentiality, the information collected about you will be given a code number so that your name and personal information can be removed from all paperwork. Information will be kept in a locked filing cabinet in the locked research laboratory or will be stored on password-protected laboratory computers. The PhD researcher, supervisors, and research assistants will have access to the data. Research data will be stored for 10 years after data collection, after which it will then be destroyed.

The results of the project may be included in the researcher's PhD thesis, and may be submitted to be published in a peer-reviewed journal. A thesis is a public document and will be available through the UC Library. Please indicate to the researcher on the consent form if you would like to receive a copy of final manuscript of the project or a basic summary of results. However, However, there may be a long delay between collecting the data and completing the final report. The principal investigator can also personally discuss the results of the study with you.

The project is being carried out as a requirement completion of a doctoral degree by Karen Ng under the supervision of Maggie-Lee Huckabee. If you have any questions about the study you can contact the investigator during work hours at (03) 364 2307 or via email at karen.ng@pg.canterbury.ac.nz.

This project has been reviewed and approved by the University of Canterbury Human Ethics Committee, and participants should address any complaints to The Chair, Human Ethics Committee, University of Canterbury, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

If you agree to participate in the study, you are asked to complete the consent form and return to the researcher.

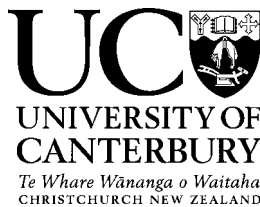
Karen Ng

Department of Communication Disorders

Telephone: +64 3 364 2307

Email: karen.ng@pg.canterbury.ac.nz

10th December 2015



Department of Communication Disorders
 Telephone: +64 3 364 2307
 Email: karen.ng@pg.canterbury.ac.nz
 10th December 2015

Consent Form

Effect of effort and bolus type on submental surface electromyographic activity during swallowing

I have been given a full explanation of this project and have had the opportunity to ask questions.

I understand what is required of me if I agree to take part in the research.

I understand that participation is voluntary and I may withdraw at any time without penalty. Withdrawal of participation will also include the withdrawal of any information I have provided should this remain practically achievable.

I understand that any information or opinions I provide will be kept confidential to the PhD researcher, supervisors, and research assistants, and that any published or reported results will not identify the participants. I understand that a thesis is a public document and will be available through the UC Library.

I understand that all data collected for the study will be kept in locked and secure facilities and/or in password-protected electronic form and will be destroyed after ten years.

I understand that I can contact the researcher Karen Ng (karen.ng@pg.canterbury.ac.nz) or her supervisor Maggie-Lee Huckabee (maggie-lee.huckabee@canterbury.ac.nz) for further information. If I have any complaints, I can contact the Chair of the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

I do not have any medical problems that may affect my swallowing.

- ☐ Optional: I would like to receive a summary of the findings.
If so, please provide email address:

By signing below, I agree with the statements above, and to participate in this research project.

Print name of participant: _____

Signature of participant: _____

Date: _____



Participant Information Sheet

Study title: Measurement of skill and strength in swallowing

Locality: University of Canterbury Rose Centre

Ethics committee ref: 15/CEN/150

Researching investigator: Karen Ng

Contact phone number: +64 (3) 364 2307

You are invited to take part in a study looking at swallowing in 3 groups of people: healthy people, people who have had a stroke, and people who have a muscle disorder. Whether or not you take part is your choice. If you don't want to take part, you don't have to give a reason, and it won't affect any care you receive. If you agree to take part, but change your mind, you can withdraw from the study at any time.

This information sheet will help you decide if you'd like to participate. It sets out why we are doing the study, what your participation would involve, what the benefits and risks to you might be, and what happens after the study ends. We will go through the information with you and answer any questions you may have. You do not have to decide today whether or not you will participate in this study. Before you decide you may want to talk about the study with others, such as family, whānau, friends, or healthcare providers. Feel free to do this. If you need an interpreter, this can and will be provided.

If you agree to take part in this study, you will be asked to sign the Consent Form on the last page. You will be given a copy of both the Information Sheet and the Consent Form to keep.

This document is 11 pages long, including the Consent Form. Please make sure you read and understand everything.

WHAT IS THE PURPOSE OF THE STUDY?

We will measure the skill and strength of muscles involved in swallowing. We want to determine whether there are any patterns of swallowing impairment within people who have trouble swallowing because of a stroke or muscle disorder, compared to healthy people. This study will give us a better understanding of how the brain controls swallowing, and what happens to swallowing when the brain is damaged. We hope to find a total of 120 participants in 14 months.

The supervising investigator is Dr. Maggie-Lee Huckabee. She is a Professor in the Department of Communication Disorders, and Director of the University of Canterbury Rose Centre for Stroke Recovery and Research. The researching investigator is Karen Ng. She is a Ph.D. student at the University of Canterbury and has a Master of Arts degree in Speech-Language Therapy.

This study has been reviewed and approved by The Health and Disability Ethics Committees (HDECs).

You can be chosen to participate if you are over 50 years old, and fall in 1 of 3 groups:

1. Healthy group: You are healthy and have never had a brain injury (e.g., stroke), head/neck injury, problems with swallowing, or a muscular disorder.

Or

2. Stroke group: You have had a stroke and have difficulty swallowing.

Or

3. Muscle disorder group: You have inclusion body myositis, oculopharyngeal muscular dystrophy, or myotonic dystrophy, and have difficulty swallowing.

WHAT WILL MY PARTICIPATION IN THE STUDY INVOLVE?

The study will take 60 – 90 minutes in a single session to complete. The research procedure consists of 2 parts.

1. Clinical swallowing evaluation

The researcher will conduct a clinical swallowing evaluation to identify any swallowing difficulties.

- a) You will complete a questionnaire about your swallowing and your health. If you don't know the answers to the questions, the researcher may ask for your permission to contact your GP for more information.
- b) The researcher will evaluate the nerves and muscles used for swallowing by asking you to make certain movements with your mouth, tongue and face (for participants with difficulty swallowing only).
- c) The researcher will assess your swallowing when you eat small amounts of food, and drink water from a cup. A video-recording will be made. The video will be viewed by the investigators only to determine results, and will be deleted on conclusion of the study.

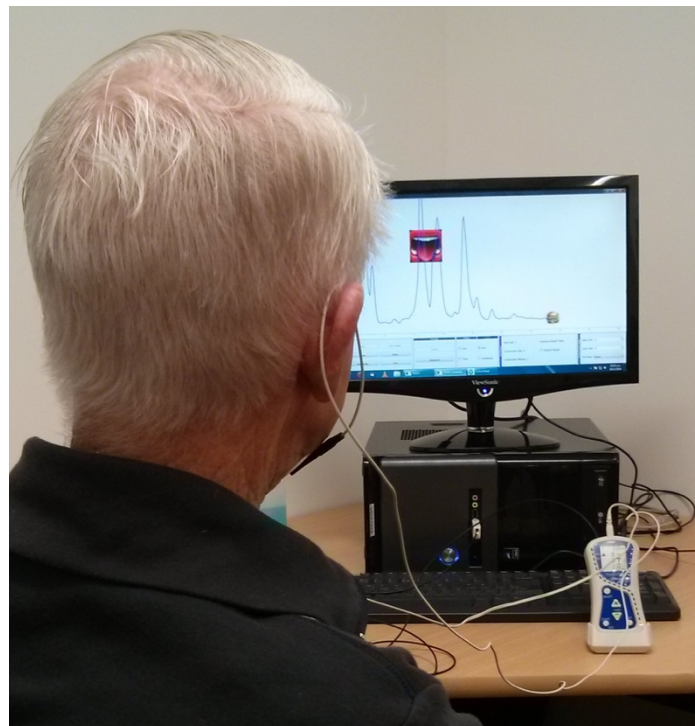
2. Strength and skill assessments

We will measure activity of the muscles used in swallowing with a sensor that attaches under your chin. The skin under the chin needs to be clean-shaven.



You will complete the following 4 tasks:

- a) Swallow as hard as you can, and swallow normally (5 times each).
- b) Jaw strength test: open your jaw against resistance (5 times).
- c) Hit a target on a computer screen by controlling the timing and force of your swallowing (10 times).
- d) Hit a target on a computer screen by controlling the timing and force of your mouth opening (10 times).



WHO PAYS FOR THE STUDY?

The University of Canterbury Rose Centre for Stroke Recovery and Research will pay for this study. You will have no costs for being a part of this study. You will be given a \$10 petrol voucher if you have travel expenses.

WHAT ARE THE POSSIBLE BENEFITS AND RISKS OF THIS STUDY?

The benefits of this study are a better understanding of your swallowing function. The jaw strength test requires you to open your jaw with maximum force against resistance, 5 times. It is possible that you may feel soreness and discomfort in your jaw and/or neck. This procedure has been used in many previous studies with no documented serious side effects. For safety reasons, people who have temporomandibular joint (TMJ) or jaw pain/disorders should not take part in this study. You will be monitored very carefully by the researchers for any possible risk during the study. In the unlikely event that you feel any pain during jaw opening, the study will be stopped immediately.

WHAT IF SOMETHING GOES WRONG?

If you were injured in this study, which is unlikely, you would be eligible to apply for compensation from ACC just as you would be if you were injured in an accident at work or at home. This does not mean that your claim will automatically be accepted. Your claim may take some time to assess. If your claim is accepted, you will receive funding to assist in your recovery. If you have private health or life insurance, you may wish to check with your insurer that taking part in this study won't affect your cover.

WHAT ARE MY RIGHTS?

Your participation is voluntary. Whether or not you take part is your choice. If you don't want to take part, you don't have to give a reason, and it won't affect the care you receive. If you do want to take part now, but change your mind later, you can pull out of the study at any time.

You have the right to access information about yourself collected as part of the study. You will be told of any new information related to the study that may have an impact on your health. The information collected about you will be given a code number so

that your name and personal information can be removed from all paperwork.

WHAT HAPPENS AFTER THE STUDY OR IF I CHANGE MY MIND?

After the study, data may be included in the investigator's PhD thesis. With your permission, data may be used in future related studies, which have been given ethical approval from the Health and Disability Ethics Committees. We may also submit results to be published in a peer-reviewed journal. However, there will be no information that could personally identify you.

Consent forms will be kept in a locked filing cabinet in the locked research laboratory or will be stored on password-protected laboratory computers. Research data will be stored for 10 years after data collection. The data will then be destroyed.

If you wish, we can give you copies of the final report. However, there may be a long delay between collecting the data and completing the final report. The principal investigator can also personally discuss the results of the study with you.

If you agree to take part in the study, but change your mind, you can withdraw from the study at any time.

WHO DO I CONTACT FOR MORE INFORMATION OR IF I HAVE CONCERNS?

If you have any questions, concerns or complaints about the study at any stage, you can contact the researchers:

Karen Ng
PhD Candidate
Phone: (03) 364 2307
Email: karen.ng@pg.canterbury.ac.nz

Dr. Maggie-Lee Huckabee
Professor
Phone: (03) 364 2042
Email: maggie-lee.huckabee@canterbury.ac.nz

If you want to talk to someone who isn't involved with the study, you can contact an independent health and disability advocate on:

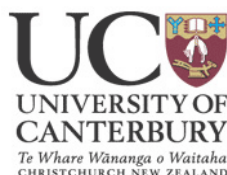
Phone: 0800 555 050
Fax: 0800 2 SUPPORT (0800 2787 7678)
Email: advocacy@hdc.org.nz

For Maori health support please contact:

Dr. Tracy Rohan
Research Consultant Maori, University of Canterbury
Phone: (03) 364 2987 ext. 45520
Email: tracy.rohan@canterbury.ac.nz

You can also contact the health and disability ethics committee (HDEC) that approved this study on:

Phone: 0800 4 ETHICS
Email: hdec@moh.govt.nz



Consent Form

If you need an INTERPRETER, please tell us.

I have read, or have had read to me in my first language, and I understand the Participant Information Sheet.

I have been given sufficient time to consider whether or not to participate in this study.

If needed, I have had the opportunity to use a legal representative, whānau/ family support or a friend to help me ask questions and understand the study.

I am satisfied with the answers I have been given regarding the study and I have a copy of this consent form and information sheet.

I understand that taking part in this study is voluntary (my choice) and that I may withdraw from the study at any time without this affecting my medical care.

I consent to the research staff collecting and processing my information, including information about my health.

I understand that my participation in this study is confidential and that no material, which could identify me personally, will be used in any reports on this study.

I agree to an approved auditor appointed by the New Zealand Health and Disability Ethic Committees, or any relevant regulatory authority or their approved representative reviewing my relevant medical records for the sole purpose of checking the accuracy of the information recorded for the study.

I understand the compensation provisions in case of injury during the study.

I know whom to contact if I have any questions about the study in general.

I understand my responsibilities as a study participant.

I consent to video-recording during part of the clinical swallowing evaluation, and for this to be viewed by the researchers for data analysis.

Optional

If I decide to withdraw from the study, I agree that the information collected about me up to the point when I withdraw may continue to be processed. Yes ☐ No ☐

I consent to the research staff contacting my general practitioner (GP) to collect information about my health. Yes ☐ No ☐

I consent to my data being used in future related studies, which have been given ethical approval from the Health and Disability Ethics Committees (HDECs). Yes ☐ No ☐

I wish to receive a summary of the results from the study. Yes ☐ No ☐

If yes, please provide email address:

Declaration by participant:

I hereby consent to take part in this study.

Participant's name:

Signature:

Date:

Declaration by member of research team:

I have given a verbal explanation of the research project to the participant, and have answered the participant's questions about it.

I believe that the participant understands the study and has given informed consent to participate.

Researcher's name:

Signature:

Date:



Participant Information Sheet

Sub-study: X-ray measurements of swallowing in people after stroke

Study title: Measurement of skill and strength in swallowing

Locality: University of Canterbury Rose Centre

Ethics committee ref: 15/CEN/150

Researching investigator: Karen Ng

Contact phone number: +64 (3) 364 2307

You are invited to take part in a sub-study looking at swallowing difficulty in people who have had a stroke.

This information sheet will help you decide if you'd like to participate. We will go through the information with you and answer any questions you may have. You do not have to decide today whether or not you will participate in this study. Before you decide you may want to talk about the study with others, such as family, whānau, friends, or healthcare providers. Feel free to do this. If you need an interpreter, this can and will be provided.

If you agree to take part in this study, you will be asked to sign the Consent Form on the last page. You will be given a copy of both the Information Sheet and the Consent Form to keep.

This document is 8 pages long, including the Consent Form. Please make sure you read and understand everything.

WHAT IS THE PURPOSE OF THE STUDY?

In the main study, we used a new test to measure the skill and strength of your muscles used for swallowing. The test showed that your stroke may have affected the strength or skill of your muscles.

Another test commonly used to evaluate swallowing is a videofluoroscopic swallowing study. This test uses an X-ray to look inside your mouth and throat, and see movement of the throat during swallowing. We want to see if the results of our new test match the results of the X-ray test.

The supervising investigator is Dr. Maggie-Lee Huckabee. She is a Professor in the Department of Communication Disorders, and Director of the University of Canterbury Rose Centre for Stroke Recovery and Research. The researching investigator is Karen Ng. She is a Ph.D. student at the University of Canterbury and has a Master of Arts degree in Speech-Language Therapy.

This study has been reviewed and approved by The Health and Disability Ethics Committees (HDECs).

You may be asked to participate in this stroke sub-study if results from the main study show that your throat muscles have decreased strength or skill.

WHAT WILL MY PARTICIPATION IN THE STUDY INVOLVE?

The study will take about 30 minutes in a single session to complete. It will take place at the University of Canterbury Rose Centre for Stroke Recovery and Research. When you are ready to start, you will be seated in a comfortable chair. We will ask you to swallow sips of water, and small bites of food. The food and water will be mixed with a small amount of barium so we can see it on the X-ray. You will hold the food or liquid in your mouth for a few seconds, and then swallow when the researcher tells you to.

WHO PAYS FOR THE STUDY?

The University of Canterbury Rose Centre for Stroke Recovery and Research will pay for this study. You will have no costs for being a part of this study. You will not be provided with payment for participating.

WHAT ARE THE POSSIBLE BENEFITS AND RISKS OF THIS STUDY?

The benefits of this study are a better understanding of your swallowing function. It can help us develop better treatments for stroke survivors with swallowing problems.

The swallowing x-ray exposes you to small amounts of radiation. However, the radiation amount is much lower than that of a regular chest x-ray. More than 40 swallowing x-rays would be needed in a year to go over the yearly radiation limit. Please let the researchers know if you are allergic to barium. Barium liquid may cause side effects, such as constipation. However, you will be drinking less barium than would be given in a regular barium swallow examination. The researchers will monitor you very carefully during this study for any changes.

WHAT IF SOMETHING GOES WRONG?

If you were injured in this study, which is unlikely, you would be eligible to apply for compensation from ACC just as you would be if you were injured in an accident at work or at home. This does not mean that your claim will automatically be accepted. Your claim may take some time to assess. If your claim is accepted, you will receive funding to assist in your recovery. If you have private health or life insurance, you may wish to check with your insurer that taking part in this study won't affect your cover.

WHAT ARE MY RIGHTS?

Your participation is voluntary. Whether or not you take part is your choice. If you don't want to take part, you don't have to give a reason, and it won't affect the care you receive. If you do want

to take part now, but change your mind later, you can pull out of the study at any time.

You have the right to access information about yourself collected as part of the study. You will be told of any new information related to the study that may have an impact on your health. The information collected about you will be given a code number so that your name and personal information can be removed from all paperwork.

WHAT HAPPENS AFTER THE STUDY OR IF I CHANGE MY MIND?

After the study, data may be included in the investigator's PhD thesis. With your permission, data may be used in future related studies, which have been given ethical approval from the Health and Disability Ethics Committees. We may also submit results to be published in a peer-reviewed journal. However, there will be no information that could personally identify you.

Consent forms will be kept in a locked filing cabinet in the locked research laboratory or will be stored on password-protected laboratory computers. Research data will be stored for 10 years after data collection. The data will then be destroyed.

If you wish, we can give you copies of the final report. However, there may be a long delay between collecting the data and completing the final report. The principal investigator can also personally discuss the results of the study with you.

If you agree to take part in the study, but change your mind, you can withdraw from the study at any time.

WHO DO I CONTACT FOR MORE INFORMATION OR IF I HAVE CONCERNS?

If you have any questions, concerns or complaints about the study at any stage, you can contact the researchers:

Karen Ng
PhD Candidate
Phone: (03) 364 2307
Email: karen.ng@pg.canterbury.ac.nz

Dr. Maggie-Lee Huckabee
Professor
Phone: (03) 364 2042
Email: maggie-lee.huckabee@canterbury.ac.nz

If you want to talk to someone who isn't involved with the study, you can contact an independent health and disability advocate on:

Phone: 0800 555 050
Fax: 0800 2 SUPPORT (0800 2787 7678)
Email: advocacy@hdc.org.nz

For Maori health support please contact:

Dr. Tracy Rohan
Research Consultant Maori, University of Canterbury
Phone: (03) 364 2987 ext. 45520
Email: tracy.rohan@canterbury.ac.nz

You can also contact the health and disability ethics committee (HDEC) that approved this study on:

Phone: 0800 4 ETHICS

Email: hdec@moh.govt.nz



Consent Form

If you need an INTERPRETER, please tell us.

I have read, or have had read to me in my first language, and I understand the Participant Information Sheet.

I have been given sufficient time to consider whether or not to participate in this study.

If needed, I have had the opportunity to use a legal representative, whānau/ family support **or** a friend to help me ask questions and understand the study.

I am satisfied with the answers I have been given regarding the study and I have a copy of this consent form and information sheet.

I understand that taking part in this study is voluntary (my choice) and that I may withdraw from the study at any time without this affecting my medical care.

I consent to the research staff collecting and processing my information, including information about my health.

I understand that my participation in this study is confidential and that no material, which could identify me personally, will be used in any reports on this study.

I agree to an approved auditor appointed by the New Zealand Health and Disability Ethic Committees, or any relevant regulatory authority or their approved representative reviewing

my relevant medical records for the sole purpose of checking the accuracy of the information recorded for the study.

I understand the compensation provisions in case of injury during the study.

I know whom to contact if I have any questions about the study in general.

I understand my responsibilities as a study participant.

Optional

If I decide to withdraw from the study, I agree that the information collected about me up to the point when I withdraw may continue to be processed.

Yes ☐ No ☐

I consent to my data being used in future related studies, which have been given ethical approval from the Health and Disability Ethics Committees (HDECs).

Yes ☐ No ☐

Declaration by participant:

I hereby consent to take part in this study.

Participant's name:

Signature:

Date:

Declaration by member of research team:

I have given a verbal explanation of the research project to the participant, and have answered the participant's questions about it.

I believe that the participant understands the study and has given informed consent to participate.

Researcher's name:

Signature:

Date:
